Effective Detection and Treatment of Amblyopia: Addressing Noncompliance

Effectieve Detectie en Behandeling van Amblyopie: Aandacht voor Therapieontrouw

Angela Mei-Lian Tjiam

Effective Detection and Treatment of Amblyopia: Addressing Noncompliance

Thesis, Erasmus University Rotterdam, The Netherlands

The research project was initiated by the department of Ophthalmology, ErasmusMC University Medical Centre Rotterdam, The Netherlands. The work described in this thesis was financially supported by ZonMW – The Netherlands Organization for Health Research and Development (grant number: #28.2689.0 (chapter 2) & #6320.0008).

The printing of this thesis was financially supported by:

ISBN: 978-94-6169-270-2

Lay-out & Printing: Optima Grafische Communicatie, Rotterdam, The Netherlands

Cover & Design: VOF Vingerling & De Bruyne

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Effectieve Detectie en Behandeling van Amblyopie:
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Proefschrift

ter verkrijging van de graad van doctor aan de
Erasmus Universiteit Rotterdam
op gezag van de rector magnificus

Prof.dr. H.G. Schmidt

en volgens besluit van het College voor Promoties.
De openbare verdediging zal plaatsvinden op

woensdag 21 november 2012 om 15:30 uur

door

Angela Mei-Lian Tjiam
geboren te Goes

ERASMUS UNIVERSITEIT ROTTERDAM

PROMOTIECOMMISSIE

Promotoren:	Prof.dr. G. van Rij
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Chapter 1

General Introduction

GENERAL INTRODUCTION

1.1 Amblyopia & Compliance, literature overview

Amblyopia (a 'lazy eye') is commonly defined as a decrease in visual acuity (sight) in either or both eyes which persists after correction of the refractive error (by wearing glasses) and / or removal of any pathological obstacle to vision (Ansons et al. 2009). In the clinical setting amblyopia is generally expressed as a loss of visual acuity, and it usually presents itself during the ophthalmological examination by the ophthalmologist or the orthoptist (Levi 2006). It is usually associated with the presence of amblyogenic factors such as strabismus (ocular misalignment causing each eye to have a different image on the fovea), a refractive error (one foveal image is more blurred than the other); or, in rare cases, deprivation of a clear retinal image (physical obstruction, e.g. infantile cataract or ptosis) (Ciuffreda 1991; Von Noorden 1967, 1985; Von Noorden et al. 2002b).

It affects approximately 3%-4% of the adult population (Attebo et al. 1998). This prevalence, however, varies in literature (0.5-5.3%) due to differences in study design, population and the examination methods used (Attebo et al. 1998; Cole 1959; Helveston 1965; Simons 1996; Theodore et al. 1946; Vinding et al. 1991; Von Noorden et al. 2002a). Nonetheless, amblyopia is the most common eye disorder in young children (Noorden von et al. 2002; Searle et al. 2002), and in people aged 20-70 it is the most common cause of monocular loss of vision (Buch et al. 2001).

Reduction of vision in amblyopic patients is due to a dysfunction of the processing of visual information in the first years of life (Holmes et al. 2006a). The etiological mechanism underlying amblyopia has been extensively studied in kittens and young monkeys. These studies have shown that electrophysiological and anatomical changes take place in the brain (i.e. reduction of neural cells in the primary visual cortex and lateral geniculate nucleus) after disruption of visual stimulus during the critical period in an animal's life (Ansons et al. 2009; Blakemore et al. 1986; Crawford et al. 1975; Crawford et al. 1989; Crawford et al. 1984; Crawford et al. 1979; Ikeda et al. 1974a, b; Moseley et al. 2002a; Smith et al. 1979; Von Noorden et al. 1975; Wiesel et al. 1963b, 1965).

Recovery of amblyopia is possible by promoting the use of the amblyopic eye with complete or partial exclusion of the better eye (Von Noorden et al. 2002c). This treatment aims to improve the visual acuity of the amblyopic eye to its optimum level, preferably to equal acuity in both eyes (Ansons et al. 2009). For the last 250 years, occlusion of the better eye by an opaque eye patch has been the mainstay of treatment of amblyopia (Loudon 2005; Webber et al. 2005). However, therapeutic regimens have lacked standardization, with the length of patching ranging from a few minutes a day to all waking hours and in

some cases treatment may last many months to years (Loudon et al. 2004; Tan et al. 2003; Webber 2007). Nonetheless, occlusion therapy with the eye patch is the most commonly used treatment method to treat amblyopia in daily practice (Bacal 2004; Doshi et al. 2007; Webber et al. 2005). Other therapeutic methods than occlusion of the better eye are optical penalization (selectively fogging the image of the non-amblyopic eye by glasses), pharmaceutical penalization (cycloplegia by the daily instillation of drops into the fornix of the non-amblyopic eye) and other non-conventional treatments, such as Red-Filter treatment, prisms and CAM treatment (Holmes et al. 2006a; Loudon 2005). Although recent studies investigated the pros and cons of occlusion therapy and the effectiveness and acceptability of the various treatment methods (Cole et al. 2001; Holmes et al. 2003; Holmes et al. 2001; Holmes et al. 2005; Holmes et al. 2006b; Scheiman et al. 2005), this thesis focuses only on the occlusion therapy with the eye patch.

Amblyopia develops only in the critical period (Von Noorden et al. 1979). Once this period is over amblyopia does not develop or amblyopia is irreversible. It is suggested - based on results from studies on form vision in monkeys - that the comparable period in man in which form vision develops and amblyopia can occur is approximately eight years after birth (Ansons et al. 2009; Stager et al. 1990; Von Noorden et al. 1979). However, the extent of the critical period in humans is matter of some debate (Fronius et al. 2009; Moseley et al. 2002a); there appears to be substantial plasticity beyond the "critical period" (Fronius et al. 2009; Levi 2006).

Early detection of Amblyopia

With this critical period in mind, treatment of amblyopia should occur in early childhood: the younger the patient the better and more rapid the response (Ansons et al. 2009). Treatment is rarely started after the age of eight years, when the visual system has matured, as it has been shown that treatment later in the critical period becomes less effective (Birch et al. 1990; Crawford et al. 1983; Epelbaum et al. 1993; Mintz-Hittner et al. 2000; Mitchell 1991).

For these reasons vision screening programs to early detect amblyopia and/or strabismus have been developed, evaluated and used in some countries. Sweden, the United Kingdom, The Netherlands, and some eastern European countries have population-based programs for child vision screening. In general, the purpose of screening is to identify a specific, unrecognized disease whereof its treatment is (more) effective when started at an early stage (Commission on chronic illness 1957; Ohlsson 2006). It is important to keep in mind that screening is not intended to be diagnostic.

There are a variety of recommendations for vision screening methods and a number of different approaches to providing the service (Carlton et al. 2008; Powell et al. 2007). This includes the age at which the child is screened, referral criteria of the screening program, and the personnel administering the test that form the screening programme (Carlton et al. 2010). In The Netherlands, childhood vision screening had been implemented in the Dutch child health screening program in the 1960s. Nowadays the Dutch vision screening consists of preverbal vision screening (corneal light reflex test, the cover-uncover test, an examination of ocular pursuit movements for both monocular and binocular conditions, inspection of the eyelids and anterior segment of the eye) and preschool screening (testing of monocular visual acuity and ocular alignment) (Loewer-Sieger et al. 1987; Van Velzen-Mol 2002; Van Velzen-Mol et al. 2006; van Velzen-Mol et al. 2003).

However, the justification of vision screening for amblyopia remains a controversial issue (Beauchamp et al. 2010; Carlton et al. 2008; IQWiG 2008; Mema et al. 2012; Snowdon et al. 1997b). Based on a systematic review, Snowdon and Stewart-Brown (1997) concluded that there is insufficient evidence to support a preschool vision screening programme and that every effort should be made to rectify this. Similarly, the IQWIG (Intitut für Qualität und Wirtschaftlichkeit im Gesundheitswesen, 2008) in Köln Germany confirmed from their review that the evidence on the benefits of vision screening is of moderate to poor quality, and that studies lacked on results on the burden of screening and overtreatment. Carlton et al. (2008) concluded that the cost-effectiveness of screening for amblyopia is dependent on the long-term utility effects of unilateral vision loss, but that evidence on such effects was limited. Likewise, Mema et al. (2012) acknowledged the need for additional population-based research in order to establish the utility of preschool vision screening in general.

Compliance in Amblyopia treatment

Despite screening and treatment, approximately a third of the affected children who have been prescribed occlusion therapy do not reach sufficient visual acuity to read properly with the amblyopic eye (Jensen et al. 1986; Rahi et al. 2002a; Vinding et al. 1991). The low degree of compliance - i.e. the degree to which a patient follows or completes a prescribed diagnostic, treatment or preventive procedure - has been reported as the major contributor to the failure of occlusion treatment (Awan et al. 2005; Awan et al. 2010; Dorey et al. 2001; Lithander et al. 1991; Loudon et al. 2006; Loudon et al. 2007a; Newsham 2000; Simmers et al. 1999; Simons et al. 1999; Stewart et al. 2005; Stewart et al. 2004b; Woodruff et al. 1994).

In general, noncompliance leads to unnecessary use of healthcare resources, as it contributes to repeated office visits, missed outpatient appointments, unsuccessful referrals,

changed treatment methods, and a longer course of illness and treatment (Mark et al. 1999; Marsh-Tootle et al. 2010; Sutton et al. 1994; Vernon et al. 1990; Williamson et al. 1995). Therefore, an increased interest on the issue of patient compliance has been received in the past decades (Adler et al. 2010; Desai et al. 2011; Matsui 2007; Osterberg et al. 2005).

However, compliance is a complex issue, partly due to difficulties in assessing it and in obtaining meaningful and accurate data. The need to objectify compliance with electronic devices has increased since Kass et al. (1986) had demonstrated in the 1980s that the objective electronically measured compliance was 30% lower than according to what was reported in the patients' dairies: with electronic measurements they showed that 76% of the pilocarpine drops for glaucoma was taken as prescribed, whereas the patients' diaries reported to have taken 97% of their medication (Kass et al. 1986a; Kass et al. 1987; Kass et al. 1986b; Norell 1980; Urquhart 1991, 1996, 1997, 1999, 2008).

Since Fielder and Moseley had developed the Occlusion Dose Monitor (ODM) in 1994, it was possible to measure compliance with occlusion therapy electronically, and therefore objectively (Moseley et al. 2002b). In previous studies with the ODM, it was found that compliance with occlusion treatment is rarely total. Compliance was 48%-57%, on average (Awan et al. 2005; Loudon et al. 2006; Stewart et al. 2004b). It was also demonstrated that treatment success is related to the level of compliance (Awan et al. 2005; Loudon et al. 2003; Simonsz et al. 1999; Stewart et al. 2002; Stewart et al. 2004b; Stewart et al. 2007b).

Factors affecting compliance

Reasons for non-compliance given by 68% of parents demonstrated poor knowledge (Newsham 2000). Good communication is important in improving compliance (Friedman et al. 2008; Tates et al. 2001; Winnick et al. 2005) and therefore essential for a good visual outcome (Matsui 2007; Simons et al. 1999; Stewart et al. 2005; Stewart et al. 2004a). Loudon et al. (2006) found that low fluency in the Dutch language, living in a low socio-economic status (SES) area, low level of education, country of origin and a low visual acuity of the amblyopic eye at the start of occlusion therapy are predictors for low compliance.

Failure in treatment due to noncompliance should be avoided as such amblyopic patients are more at risk for bilateral visual impairment after losing visual function in the better eye due to an eye trauma, glaucoma, macular degeneration or cataract (Holmes et al. 2006a; Rahi et al. 2002a; van Leeuwen et al. 2007). In one study the risk that an amblyopic patient will lose the function of the better eye later in life was estimated at 0.175% (Tommila

et al. 1981). This will subsequently result in job losses, an increased morbidity and social isolation (Chua et al. 2004; Fronius et al. 2005; Rahi et al. 2002a). In addition, a decrease in quality of life in adulthood among insufficiently treated amblyopic patients has been described (Van de Graaf et al. 2004; Van de Graaf et al. 2007).

Improving compliance

Several studies on glaucoma, asthma, HIV or diabetes medication have been conducted to investigate how to improve patients' compliance. Examples to improve compliance which were investigated are: information programmes, personal mentoring by pharmacists, reviewed current barriers and possible solutions with a study coordinator, phone call reminders, audible and visible remainders activated on their Dosing Aid devices, educational videotapes, educational books (Costello et al. 2004; Haynes et al. 2008; Holzheimer et al. 1998; Indinnimeo et al. 2009; Schedlbauer et al. 2010).

Studies on improving compliance are mostly focused on adult patients; less is known about improving compliance in children. With regard to improving compliance with occlusion therapy, few studies reported positive results by giving written information to the parents (Newsham 2002). In 2006, a randomized control trial (Loudon et al. 2006) reported on the effectiveness of an educational cartoon ("*Het Plakboek*") that improved electronically measured compliance with occlusion therapy. This cartoon was made by José Vingerling and Gerard de Bruyne, two artists who are specialised in art for sick children. It focused primarily on the children, and consisted of a cartoon story that explained, without words, why children should wear the eye patch, and was accompanied with a reward calendar with stickers, and, for the parents, an information leaflet in six languages (Dutch, English, French, German, Turkish and Arabic). In children who used the cartoon, mean electronically measured compliance was 78%, against 56% who did not. Also, it was demonstrated that only two percent of the children who used the cartoon patched less than 30% of the prescribed occlusion time against 15% in the controls (Loudon et al. 2006).

Although the effectiveness of this educational cartoon was demonstrated, adoption of valuable insights, procedures or products does not generally take place easily or completely. It may well be that the educational cartoon and other compliance-enhancing measures cannot be implemented in daily practice. In literature, it has been described that there is a gap between available evidence and current practice in health care (Glasgow et al. 2003; Grimshaw et al. 2004; Grimshaw et al. 2002; Grol et al. 1999; Horner et al. 2004; Stetler et al. 2008). Implementation studies were conducted to identify problems and strategies were developed to process evidence based results into real practice (Cabana et al. 1999; Grol et al. 2005).

1.2 Problem statement & Aims of this thesis

In general, noncompliance with something, for example a law, treaty, or agreement means not doing what you are required or expected to do. Within healthcare, noncompliance may not only limit the effectiveness of curative care, but also that of preventive care.

In 2001, it was found in the Rotterdam AMblyopia Screening Effectiveness Study (RAMSES) that one third of all children with a positive screening test result in population-based vision screening were not seen by an orthoptist or ophthalmologist (Juttmann 2001). For the first time, the serious effect of unsuccessful referral after a positive screening test on screening for amblyopia was demonstrated.

Five years later, Loudon et al. (2006) demonstrated that compliance with occlusion therapy for amblyopia, objectified with electronic measurements, was 57%, on average – i.e. children wore the patch only 57% of the time prescribed by the orthoptist. This was an unexpectedly low finding. They showed that this low compliance was related to the parental fluency in the native language, country of origin and level of level of education, and that compliance could be enhanced by the use of an educational cartoon that explains to the child why it should wear the eye patch.

Until that time, in the field of ophthalmology, the magnitude of the issue on noncompliance was little known. Electronic measurement of compliance had only been performed in eye drops for treatment of glaucoma. There was limited knowledge on the impact of noncompliance on treatment and treatment outcome, causes for noncompliant behaviour, the extent of attention to noncompliance among healthcare professionals and how professionals cope with noncompliance, and whether and how compliance could be enhanced.

Given these findings, the aim of this thesis was to study conditions, causes, impact, awareness, handling and improving of noncompliance, which limits the effectiveness of detection and treatment of amblyopia. Based on these problem statements, the following research questions were addressed:

What is the contribution of vision screening to detect amblyopia in children between 0 and 7 years of age? (**Chapter 2**)

To what extent is unsuccessful referral after a positive vision-screening test explained by noncompliance with referral? (**Chapter 3**)

How do orthoptists cope with noncompliance? (**Chapter 4**)

Is compliance influenced by size, colour, elasticity, and marketing policy of different brands of eye patches? (**Chapter 5**)

Do immigrant-related determinants, other than country of origin, fluency in Dutch and level of education, influence compliance? In other words, do socio-cultural, psychological and integration into Dutch society affect compliance with occlusion therapy? (**Chapter 6**)

What is the effectiveness of the educational cartoon in improving compliance, in terms of electronically measured compliance with occlusion therapy, outpatient attendance rates, visual acuity outcome, and the duration of occlusion therapy? (**Chapter 7**)

What part of the educational cartoon: the cartoon story, the calendar with reward stickers or the parental information sheet, played the greatest part in the increase in compliance with occlusion therapy? (**Chapter 8**)

Can compliance-enhancing measures, such as the educational cartoon, be adopted by orthoptists, be implemented in daily practice, and be distributed to amblyopic children nationwide? Can attitude, awareness and actions to deal with noncompliance improve among orthoptists? Can education on compliance be integrated into the basic training for student orthoptists and continuing training for working orthoptists? (**Chapter 9**)

Chapter 2

Rotterdam AMblyopia Screening Effectiveness Study: detection and causes of amblyopia in a large birth cohort

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Invest Ophthalmol Vis Sci. 2010; 51(7): 3476-84

ABSTRACT

Purpose. The Dutch population-based child health monitoring program includes regular preverbal (age range, 1–24 months) and preschool (age range, 36–72 months) vision screening. This study is on the contribution of an organized vision screening program to the detection of amblyopia.

Methods. A 7-year birth cohort study of 4624 children was started in 1996/1997 in Rotterdam. Vision screening data were obtained from the child screening centres. Treating orthoptists working at the regional ophthalmology departments provided information about diagnosis and treatment. The diagnosis was reviewed by two experts. The parents provided additional information on their child's eye history through written questionnaires and telephone interviews. At age 7 years, the children underwent a final examination by the study orthoptists.

Results. Of the 3897 children still living in Rotterdam by 2004, 2964 (76.1%) underwent the final examination. Amblyopia was diagnosed in 100 (3.4%) of these (95% CI, 2.7–4.0). At age 7, 23% had visual acuity >0.3 logMAR. Amblyopia was caused by refractive error (n = 42), strabismus (n = 19), combined-mechanism (n = 30), deprivation (n = 7), or unknown (n = 2). Eighty-three amblyopia cases had been detected before age 7. Amblyopia detection followed positive results in vision screening in 56 children, either preverbal (n = 15) or preschool (n = 41). Twenty-six other amblyopes were self-referred (n = 12, before a first positive screening test), especially strabismic or combined-mechanism amblyopia; data were uncertain for one other positively screened amblyopic child. Amblyopia remained undetected until age 7 due to unsuccessful referral (n = 4, three with visual acuity >0.3 logMAR at age 7) or false-negative screening (n = 13).

Conclusion. Most cases of amblyopia were detected by vision screening with visual acuity measurement. Preverbal screening contributed little to the detection of refractive amblyopia.

INTRODUCTION

Amblyopia affects approximately 3% of the adult population (Attebo et al. 1998). It is commonly defined as a unilateral or bilateral vision reduction due to a dysfunction of the processing of visual information in the first years of life (Holmes et al. 2006a). Most cases can be treated effectively by occlusion therapy before age six. The purpose of child vision screening is to prevent bilateral visual impairment in adult life by the early detection and treatment of amblyopia and other disorders of vision. If treatment fails, adult eye conditions, such as eye trauma, glaucoma, macular degeneration, or cataract, may cause loss of visual function in the better eye in later life, resulting in bilateral visual impairment (Rahi et al. 2002a; van Leeuwen et al. 2007). Sweden, the United Kingdom, The Netherlands, and some eastern European countries have population-based programs for child vision screening. Assessment of visual acuity at age 4 (preschool screening) is most common. In some countries, including Canada and The Netherlands, screening of visual function in infants and young children is also applied (preverbal screening) (Canadian Paediatric Society 2009; Van Velzen-Mol 2002). There is, however, inconclusive evidence of the overall and cost effectiveness of child vision screening (Carlton et al. 2008; IQWiG 2008).

In The Netherlands, a health screening program for mother and child was initiated in the early 1900s. Regular child vision screening has been part of this Dutch child health screening program since the 1960s. Initially, vision screening consisted of inspection, testing of monocular visual acuity, ocular alignment, and stereo acuity in children 3 years of age or older (preschool/school screening). In the 1980s, an additional method of screening visual function in infants and toddlers (preverbal screening) was implemented: the VOV method (Vroegtijdige Onderkenning Visuele stoornissen, or Early Detection of Visual Disorders) (Loewer-Sieger et al. 1987). The VOV examination includes the corneal light reflex, cover–uncover test, observation of ocular pursuit movements, inspection of the cornea and pupil, and a pupillary light reflex test.

In 1996, we started a follow-up study of a birth cohort of 4624 children in actual screening practice in the city of Rotterdam: the Rotterdam AMblyopia Screening Effectiveness Study (RAMSES). The purpose of this study was to determine the sensitivity, specificity, and effectiveness of the Dutch child vision screening program up to age 7. The baseline characteristics of vision screening activities in children aged 0 to 2 years have been published (Juttmann 2001). In the present study, we analyzed clinical and screening data to describe amblyopia detection in children between 0 and 7 years of age and the contribution of preverbal and preschool vision screening to the detection of amblyopia.

METHODS

Design

Our study was a population-based, prospective, birth-cohort study. It was an observational study, our main objective being to evaluate the current practice of vision screening, referral, and follow-up.

Child Vision Screening in The Netherlands

The Netherlands has a nationwide health screening program for mother and child. The Child Health Care System provides preventive health care to all children aged 0 to 19 years living in The Netherlands and includes immunization and monitoring of growth and development. Participation in this free program is high, since 1997 ranging from 99% to 100% for infants to 72% to 87% for school children (Statistics Netherlands 2010). Child health screening is performed by nurses and screening physicians who specialize in preventive child health care, including child vision screening. Child vision screening is performed according to national guidelines (Van Velzen-Mol 2002) and consists of a series of consecutive screening examinations between the ages of 0 and 6. Until age 4, children are screened at one of the Child Health Centres (CHCs). From the age of 4, they are monitored by the municipal Public Health Service (PHS).

Preverbal Screening

At the CHCs, the VOV method is applied during regular visits at ages 1 to 2, 3 to 4, 6 to 9, 14, and 24 months. The VOV method consists of the corneal light reflex test and the cover–uncover test to detect the presence of strabismus; an examination of ocular pursuit movements for both monocular and binocular conditions to obtain a gross estimate of visual acuity; inspection of the eyelids and anterior segment of the eye, in particular the cornea; inspection of the colour and shape of the pupil; and testing of the pupillary light reflex.

Preschool/School Screening: Visual Acuity Measurements

At age 36 months, monocular visual acuity is tested by means of the Amsterdam picture chart (Amsterdamse Plaatjeskaart; APK). The APK is not logarithmic and does not use standardized optotypes. The cooperation of 3-year-old children with the APK is very good, making the test popular with staff at CHCs. At age 45 months, monocular visual acuity is measured at a CHC by means of the Landolt-C chart. If the child does not seem to understand the Landolt-C testing, the APK is used instead. A final standard vision examination with the Landolt-C chart is performed between 5 and 6 years of age at the PHS.

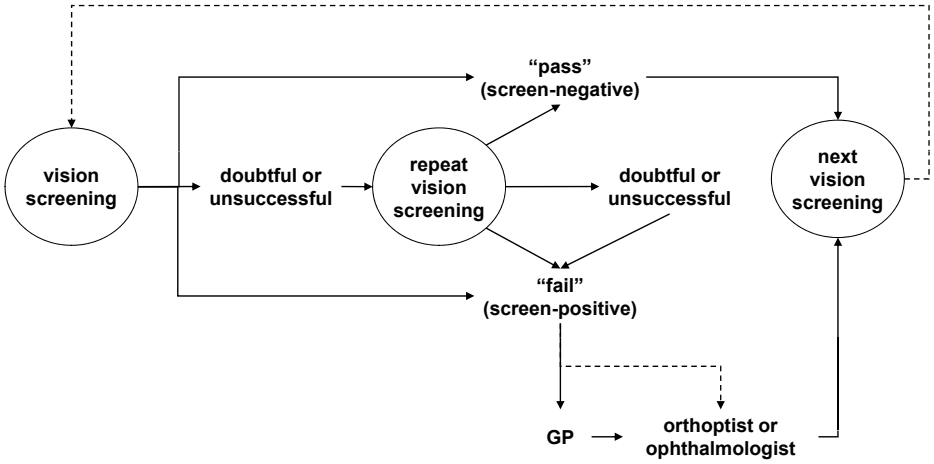

Figure 2.1) Recall (repeat vision screening) and referral policy in the Dutch vision screening program.

Referral Strategy

In case of an abnormal, that is, positive screening test result, the child is referred to an ophthalmologist or orthoptist for further assessment (usually via the general practitioner; Fig. 2.1). The VOV test result is deemed positive if one or more items are abnormal. In case of doubt about the results—for instance, an uncooperative child—the test should be repeated within 6 weeks. The referral and recall criteria for visual acuity measurements are presented in Appendix I. In The Netherlands, nearly all orthoptists work in ophthalmology departments in hospitals.

Setting

We performed our study in Rotterdam, the second largest city in The Netherlands. The city itself has approximately 592,660 inhabitants (as of January 1, 2000) and is located in an urban area (Rijnmond) with >1.2 million inhabitants. Forty percent of inhabitants in Rotterdam have a non-Dutch ethnic background (Statistics Netherlands 2008). In Rotterdam, child health screening is offered at 27 CHCs and 20 offices of the Rotterdam PHS.

Cohort

The cohort consisted of 4624 children living in Rotterdam who were born between September 16, 1996, and May 15, 1997. The children were enrolled at the 9-month visit to the CHC after the parents had given their oral informed consent. All children were offered the regular Dutch health screening program for mother and child, including vision screening.

Data Collection

Vision screening data and clinical data were prospectively collected by child health care staff and treating orthoptists, respectively, and reported to the study centre at Erasmus Medical Centre (MC).

Screening data were provided by the CHCs (0–4 years) and the Rotterdam PHS (4 years or older). If a child had visited an orthoptist or ophthalmologist before the age of 9 months, the study centre was informed as to whether the visit had been the result of previous vision screening examinations.

Treating orthoptists working at the eight ophthalmology departments in Rotterdam and its suburban areas, Capelle aan den IJssel and Spijkenisse, provided clinical orthoptic and ophthalmic data to the study centre. They filled out a standard form for each visit of a child in the birth cohort with questions concerning the diagnostic tests and treatment. The treating orthoptists were also asked to indicate whether the child (possibly) had amblyopia or any other eye disorder, and, if amblyopia was suspected, whether it was due to strabismus, a refractive error, or any other amblyogenic factor. In the summer of 2002, the study centre sent a list of all the children who had been referred after screening, but from whom no clinical data were received, to the orthoptists at the ophthalmology departments to obtain as complete a record of orthoptic and ophthalmic data as possible.

The study centre sent additional questionnaires to all parents about their children's eye history, including questions about vision screening and visits to an ophthalmology department in 2004. These questionnaires provided additional information on the follow-up of any positive screening results and revealed that some children visited other than the eight regional ophthalmology departments participating in our study, or that the general practitioner had decided that further referral to an ophthalmology department was not necessary. In 2006, after the final examination of the study, parents of children with positive vision screening tests, but without known ophthalmic or orthoptic follow-up, were contacted by additional phone or home visits. These visits are described in Chapter 3 of this thesis.

In 2004, children underwent the final examination of the study. They were approached through their schools. A team of seven independent study orthoptists recruited from outside Rotterdam visited 174 schools to assess visual acuity with the Landolt-C test; to assess ocular alignment, eye motility, and stereopsis with the TNO random dot stereotest; to check eye convergence; and to inspect the outer aspects of the eye. Children with visual acuity of 0.2 logMAR or less in one eye, a difference of 2 logMAR lines or more, manifest strabismus, or decreased stereopsis (≥240 sec/arc) were invited for an extensive orthoptic

evaluation by a team of study orthoptists and an ophthalmologist (HJS). This additional eye examination consisted of stereo testing, examination of eye position and eye movements, visual acuity measurement with a Snellen chart, retinoscopy (under cycloplegia if deemed necessary), and examination of the anterior chamber and fundus of the eye.

Main Outcome Measures

The diagnosis of interest was amblyopia and its underlying cause—that is, strabismus, refractive errors (anisometropia, isoametropia, or astigmatism), combined mechanism (a combination of strabismus and anisometropia), or deprivation (organic eye disorder). Amblyopia was defined as an interocular acuity difference of 2 logMAR lines or more or a bilateral visual acuity >0.2 logMAR in the presence of an amblyogenic factor—that is, strabismus without alternating fixation, anisometropia, astigmatism, severe myopia, or stimulus deprivation.

The following diagnostic sources were used:

1) The diagnostic information provided by the treating orthoptists. Two experts (VKL and HJS) reviewed the diagnosis proposed by the orthoptists in hindsight, with the aid of the additional data obtained later in the study. Both experts were blinded to the screening results. The diagnosis of (possible) amblyopia was upheld on expert review if it was likely that the difference in interocular visual acuity had been 2 logMAR lines or more from the start of treatment, if no alternating fixation was present, and if amblyogenic factors were identified. In cases of insufficient clinical data due to noncompliance with follow-up appointments or due to missing visual acuity measurements or cycloplegic refraction data, an expert diagnosis was made based on the results of the final examination.
2) The vision data collected in the final study examination.

Children without amblyopia were classified according to the primary disorder diagnosed by the treating orthoptists: manifest or latent strabismus; refractive errors, including (bilateral) hypermetropia, (bilateral) astigmatism, and myopia; other eye disorders, including conjunctivitis, ptosis, lacrimal disorders, and retina or optic nerve disorder; or no eye disorder. We set the threshold for insufficiently treated or residual amblyopia at a visual acuity in the amblyopic eye worse than 0.3 logMAR at age 7 years.

Statistical Analysis

All data were entered into a database (Access; Microsoft, Redmond, WA). Relevant data were converted to a statistical analysis file (SPSS ver. 15.0 for Windows; SPSS, Chicago, IL) to enable calculation of frequencies, means, and standard deviations.

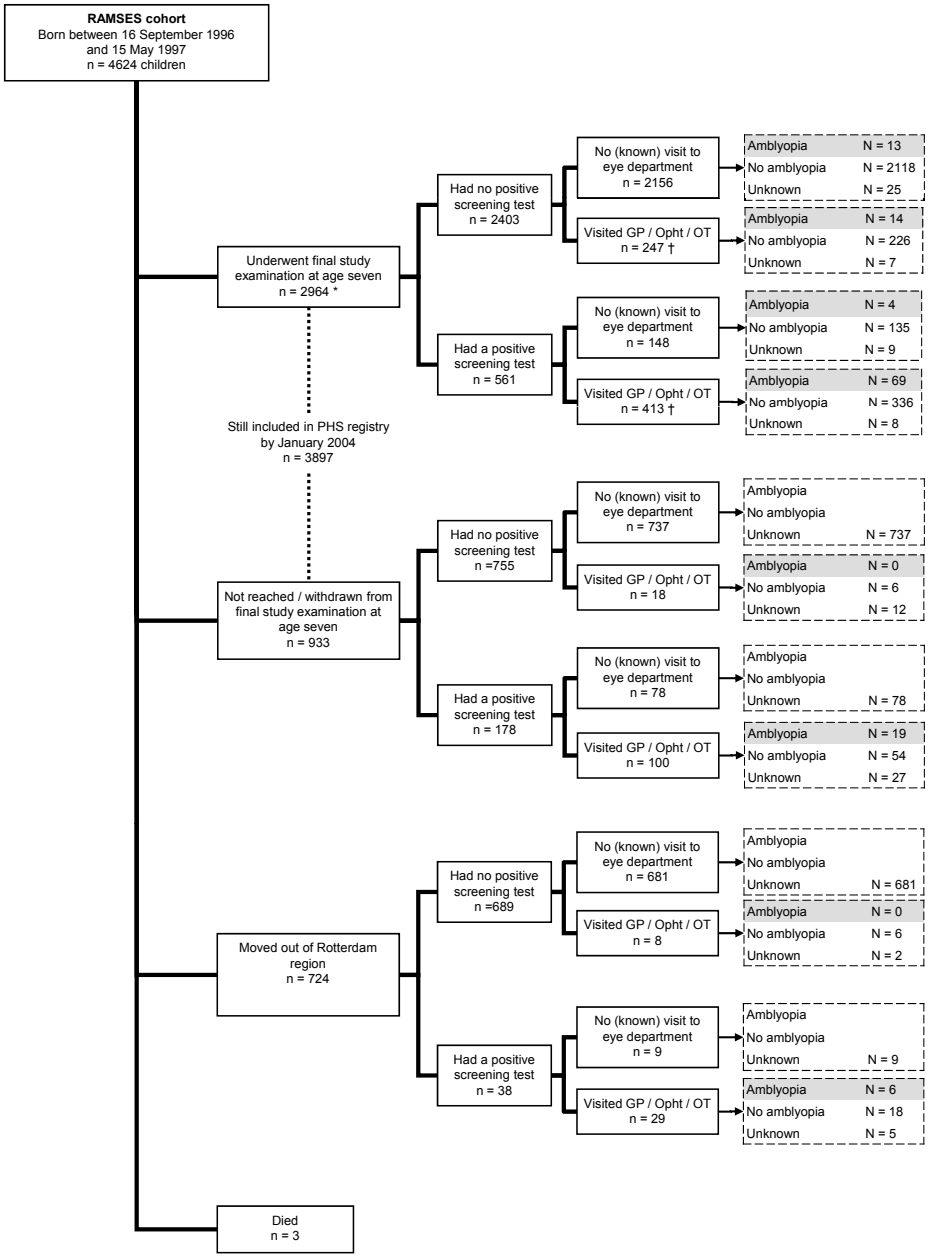

Figure 2.2) Screening result, orthoptic, or ophthalmic follow-up and amblyopia prevalence in a cohort of 4624 children between ages 0 and 7. By January 2004, 3897 of the 4624 children were still living in Rotterdam. Of these, 2964 underwent the final study examination at age 7 (the first branch of the diagram), whereas 933 other children were not examined at age 7 (the second branch). * On expert review of all data, a diagnosis of amblyopia was made in 100 of the 2964 children examined at age 7. † Of 2964 children, 660, with or without a positive vision screening test, had visited a general practitioner or an orthoptist or ophthalmologist at least once, as reported by the ophthalmology departments (384 children; for these children, clinical data were available) or by the parents (276 children).

RESULTS

By January 2004, 3897 (84.3%) of 4624 children were still registered at the Rotterdam PHS. Seven hundred twenty-four other children had moved out of Rotterdam, and three children had died (Fig. 2.2). Of the 3897 children, 933 (23.9%) were not examined at age 7; no record of which schools they were attending or their schools were outside the Rotterdam region (n = 263); their schools did not cooperate (n = 242); seemed to have moved out of Rotterdam (n = 225); or was absent at the day of the examination (n = 179); or their parents did not give permission for their child to take part (n = 14). Another ten children who were mentally retarded were excluded from the final analysis, because it was not possible to assess their visual acuity using the Landolt-C test.

For the present analysis, we used the results of 2964 (76.1%) of 3897 children who underwent the final examination at age 7. The CHCs and the PHS had reported any positive vision screening test at least once for 561 (18.9%) of the 2964 children (95% CI, 17.5–20.3) before age 6.5 years (Fig. 2.2): 126 children had their first positive vision screening at the preverbal screening and 435 at the preschool screening. The proportion of children with positive screening results did not differ between those who underwent the final examination and those who did not (X^2 test; P = 0.9).

Six hundred sixty (22.3%) of the 2964 children (95% CI, 20.8 – 23.8), whether or not they had a vision screening test with a positive result, had visited a general practitioner or an orthoptist or ophthalmologist at least once, as reported by the ophthalmology departments (384 children) or the parents (276 children) (Table 2.1).

Clinical Follow-up

Clinical data were available for 384 of the 2964 children, for whom the participating ophthalmology departments confirmed the visits to an orthoptist or ophthalmologist (see Table 2.1). The mean presenting age was 42.3 months (SD 23.0). Fifty-three of the 384 children had visited the ophthalmology department without having a positive result in the vision screening test (mean presenting age: 28.5 months, SD 27.1). The remaining 331 children had had a positive test result at least once, of whom 44 had visited the ophthalmology department before the positive screening (mean presenting age: 23.4 months, SD 16.3) and 274 after the positive screening (47.8 months, SD 20.2). The relation with the first positive screening test was unclear in the remaining 13 of the 331 children.

The most frequent diagnosis was a refractive error without amblyopia (86 [22.4%] of the children). The error was mostly hypermetropia, whereas myopia was relatively rare in children aged 0 to 7 years. The diagnosis of amblyopia was the second most frequent

Table 2.1) The diagnosis in 2964 children, according to their screening history and visit to an ophthalmology department between age 0-7

	Diagnosis *			
	Amblyopia (successfully treated or residual) n (%)	**No amblyopia** n (%)	**Unknown** n (%)	**Total †** n
Without any positive vision screening test				
No visit to ophthalmology department	13† (0.8)	1553 (99)	3 (0.2)	1569
Any visit to ophthalmology department (according to the department)	9 (17)	44 (83)	-	53
Any visit to ophthalmology department (according to parents)	5 (2.6)	182 (94)	7 (3.6)	194
Unknown visit to ophthalmology department	-	565 (96)	22 (3.7)	587
With any positive vision screening test				
No visit to ophthalmology department	4 (3.3)	115 (94)	3 (2.5)	122
Any visit to ophthalmology department (according to the department)	66 (20)	261 (79)	4 (1.2)	331
Any visit to ophthalmology department (according to parents)	3 (3.7)	75 (91)	4 (4.9)	82
Unknown visit to ophthalmology department	-	20 (77)	6 (23)	26
Total	100 (3.4)	2815 (95)	49 (1.7)	2964

* Diagnosis upon expert review of clinical information provided by the treating orthoptists working at eight regional ophthalmology departments. If clinical data were not available or insufficient, data from the final examination were reviewed to assess whether the child had had amblyopia at the time of the visits to the ophthalmology department.
† These 13 cases included six cases of non-compliance with recall or successive screening appointments and seven cases of false-negative screening. The diagnosis of amblyopia was made when the children were age 7, at the final examination of the study.

primary diagnosis (75 [19.5%] children). Seventy-one (18.5%) of the 384 children had no eye disorder. Figure 2.3 provides a more detailed overview of the primary diagnoses and how they are related to screening.

Occlusion therapy had been prescribed to 86 (22.4%) of the 384 children; 22 children received occlusion therapy only, and 64 had been prescribed glasses as well. A diagnosis of amblyopia was upheld on expert review for 61 (83%) of the 86 children who underwent occlusion therapy. One hundred three (26.8%) of the 384 children had been prescribed

glasses only. Ten (2.6%) children received treatment other than occlusion therapy or glasses, whereas no treatment had yet been prescribed or it had not been initiated at that point for 140 (36.5%) children (treatment data unknown for 45 children).

Amblyopia Prevalence

On expert review of all data, a diagnosis of amblyopia was made in 100 of the 2964 children (see also Fig. 2.1), resulting in a cumulative incidence of amblyopia of 3.4% (95% CI, 2.7–4.0) in children aged 0 to 7.

Refractive amblyopia was most frequent (n = 42 children), followed by combined-mechanism amblyopia (n = 30), strabismic amblyopia (n = 19), and deprivation amblyopia(n = 7) (unknown type [n = 2]). Of these 100 amblyopic children, 83 had visited an orthoptist or ophthalmologist according to the ophthalmology departments (n = 75) or the parents (n = 8) before age 7. Sixty-nine of these had had a positive screening test. The other 17 amblyopic children had no (known) visit to an ophthalmology department, although 4 of them had had a positive vision screening at least once. The mean presenting age was 27.2 months (SD 19.2) for strabismic amblyopia, 29.8 months (SD 19.4) for combined-mechanism amblyopia, 36.3 months (SD 29.5) for deprivation amblyopia or unknown type, and 54.1 months (SD 11.0) for refractive amblyopia.

Figure 2.4 shows the distribution of the detection of the different amblyopia types in time and in relation to screening. The points under the diagonal represent the cases of amblyopia that were detected before any positive screening test—that is, were not identified by screening. Above the diagonal are the cases of amblyopia that were detected after a positive vision screening test.

Fifty-six (56%) of 100 cases with amblyopia were detected due to the vision screening program; this proportion did not differ significantly between amblyopia caused by strabismus (12/19 cases) and refractive amblyopia (21/42 cases; X^2 test, P = 0.4). For one other child with a positive vision screening test, it was unclear whether the visit to an ophthalmology department followed the positive test. Twenty-six other cases were detected at the parents' own initiative, although 12 of these had a first positive result in a vision screening test later on. The remaining 17 children received a diagnosis only at the final study examination. Reasons for this late detection were unsuccessful follow-up after a positive vision screening test (n = 4 children) or false-negative screening (n = 13). The 13 children with false-negative screening results had different screening histories. The false-negative results were due to parental noncompliance with early recall or with successive screening examinations (n = 6) or to false-negative visual acuity measurements at preschool screening (n = 7).

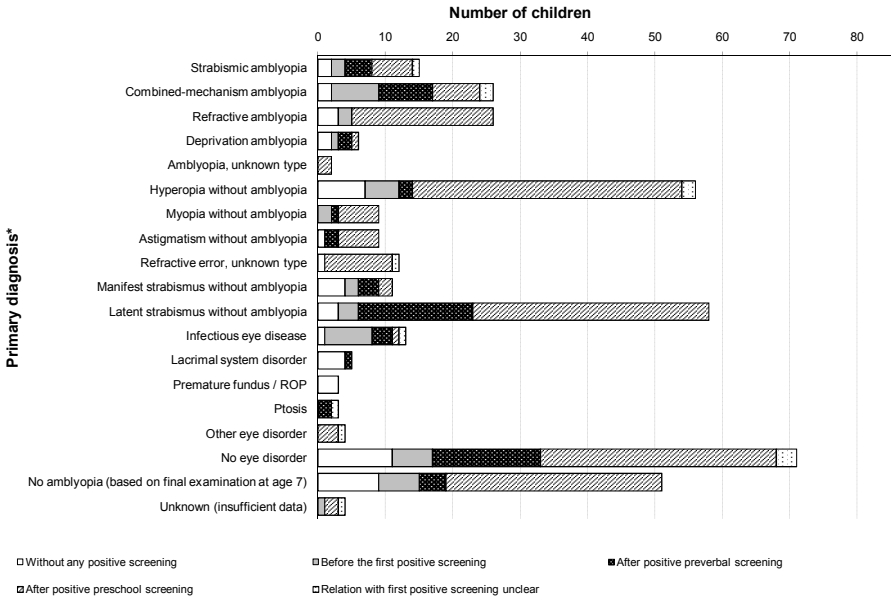

Figure 2.3) The primary diagnosis and relation to vision screening in 384 children who visited an ophthalmology department between ages 0 and 7. * Diagnosis on expert review of clinical information provided by the treating orthoptists. If clinical data were not available or were insufficient, data from the final examination were reviewed to assess whether the child had amblyopia at the time of the visits to the ophthalmology department. Orthoptists provided clinical data for 384 children in our screening cohort who had visited an ophthalmology department between ages 0 and 7 years. For 276 other children, no clinical data were received, although they had visited an orthoptist or ophthalmologist at least once, as reported by their parents (Table 2.1).

Treatment data were available for 74 of the 83 children with amblyopia who had visited an ophthalmology centre at least once before age 7. Sixty-four of them received occlusion therapy (mean age at initiation: 45.1 months, SD 22.9), and 64 received glasses (mean age at initiation: 48.3 months, SD 20.1). Fifty-five of these children received both glasses and occlusion therapy, of whom 24 were prescribed occlusion therapy first (mean interval: 11.1 months), 16 children were prescribed glasses first (mean interval: 19.5 months), and 3 were prescribed occlusion therapy and glasses at the same time (data available for 43/55 children receiving both glasses and occlusion therapy).

Figure 2.5 presents the visual acuity at age 7 of the 100 children with amblyopia. Twenty-three of the 100 children with amblyopia had visual acuity >0.3 logMAR in the worse eye at age 7. This group included 9 children with strabismic or combined-mechanism amblyopia (including 2 with untreated amblyopia), 3 with deprivation amblyopia, and 11 with refractive amblyopia (including 6 with untreated amblyopia).

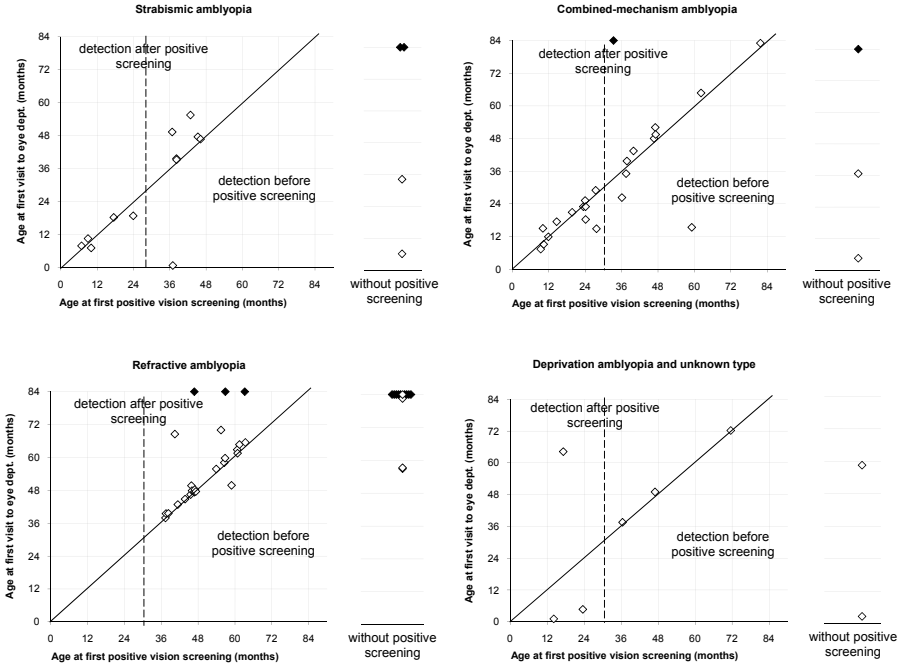

Figure 2.4) The age of amblyopia detection in relation with the first positive vision screening test. Below the diagonal: the cases of amblyopia that were detected before any positive vision screening test—that is, were not identified by screening. Above the diagonal: the amblyopia cases that were detected after a positive vision screening test. For each type of amblyopia, the children without a positive vision screening test are presented at the right. (⓿) Cases that were detected at the final study examination: 2 cases of strabismic amblyopia, 2 cases of combined-mechanism amblyopia, and 13 cases of refractive amblyopia.

DISCUSSION

We investigated the contribution of a child vision screening program to the detection of amblyopia in a large prospective birth-cohort study. We found that half of the cases of amblyopia in our cohort were detected as the result of a positive vision screening test. About one quarter of amblyopic children did not directly profit from a positive screening result because of earlier self-referral to an ophthalmology department, or because of unsuccessful referral. In the remaining quarter, the amblyopic child never had a positive result in vision screening, although half of these had received a diagnosis before age 7 after self-referral.

Preschool visual acuity measurements from age 3, in particular, played an important role in the detection of amblyopia, especially of refractive amblyopia. Cases of strabismic or

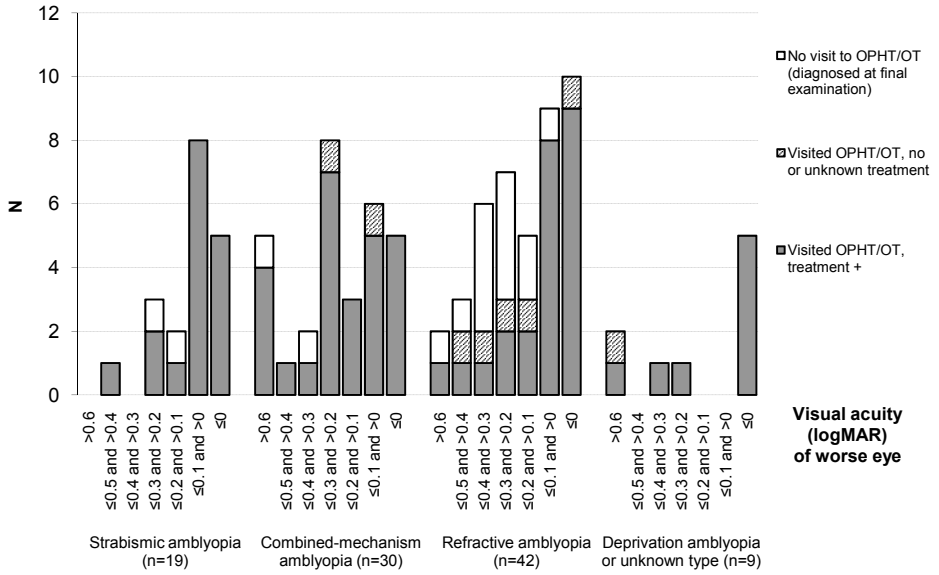

Figure 2.5) Visual acuity in the worse eye at age 7 in 100 children with amblyopia of the 2964 children who underwent the final examination of the study. Of all 100 children, 83 had visited an ophthalmology department at least once: of these, 75 had been prescribed treatment (visited OPHT/OT; treatment +), whereas no treatment data were available in 8 other cases (visited OPHT/OT, no or unknown treatment). The 17 children who had not visited an ophthalmology department between ages 0 to 7 were classified as No visit to OPHT/OT (diagnosed at final examination).

combined-mechanism amblyopia were relatively more likely to be self-referred than were those of refractive amblyopia.

Ours is the first birth-cohort study of this size on amblyopia. We were able to observe almost 3000 children from birth until age 7. There are, however, several limitations to our study. First, the provision of data was partly dependent on the attentiveness of the professionals working at the child screening centres or ophthalmology departments. We triangulated data from different sources to overcome any incomplete data. Screening and clinical data have been crosschecked and completed by parental information, and diagnostic decisions have been supported by expert review. We found that, according to parental reports, more children had visited an orthoptist or ophthalmologist than were reported by the ophthalmology departments. Part of this discrepancy can be explained by the fact that children had visited other than the participating ophthalmology departments. All children, however, were evaluated at the final study examination, and amblyopia, if present, was diagnosed at that stage. A second weakness of our study may be that in the first phase of the final examination, involving 2964 children, clinical refraction was not measured. Measuring refraction with children under cycloplegia, however, was not feasible in a population-based study of this magnitude. In the secondary phase of our

study, clinical refraction was measured with the children under cycloplegia if deemed necessary to diagnose amblyopia.

About three of four children with amblyopia attending one of the eight ophthalmology departments in our study had visited the ophthalmology department after a positive vision screening test. The visits to the outpatient ophthalmology department had not been initiated by a positive screening in the remaining quarter, although some of these children were screened positively at one of the subsequent screening examinations. The children who visited the orthoptist or ophthalmologist before or without any positive screening test were, in general, younger than 3 years of age at the first visit, and strabismic or combined-mechanism amblyopia was more common in this group than was refractive amblyopia.

We found that occlusion therapy, with or without glasses, had been prescribed to one of five children visiting the ophthalmology departments—mainly for those who had strabismic or combined-mechanism amblyopia. Orthoptists, however, had initiated occlusion therapy in some children with strabismus in whom, in hindsight, amblyopia could not be confirmed, considering all successive hospital visits. For instance, occlusion therapy could have been stopped shortly after its initiation in cases of alternating esotropia. In another Dutch study, the diagnosis of amblyopia could not be confirmed in hindsight in 7% of patients who had been prescribed occlusion therapy 30 to 35 years earlier (Simonsz-Toth et al. 2007). In our study, this percentage was 17%, but children in our study were, on average, more than 2 years younger at the start of amblyopia treatment.

To be on the safe side, orthoptists may initiate occlusion therapy in children with an uncertain diagnosis of amblyopia, because the course of treated amblyopia between termination of treatment around age 8 and adulthood varies. Visual acuity will increase slightly in most amblyopic patients, but may decrease in patients with combined-mechanism amblyopia or with increasing anisometropia (Simonsz-Toth et al. 2007).

The cumulative incidence of cases of amblyopia in this study is estimated at 3.4%. This rate is in line with previous estimates of amblyopia prevalence, ranging from 0.02% to 5.3% (Robaei et al. 2006). Because of the different definitions of amblyopia, results cannot be easily compared.

In our study, one quarter of amblyopic children—that is, 0.8% of the total population—had visual acuity in the worse eye of >0.3 logMAR at age 7. In a sample of 6-year-old Australian children, this percentage was 0.7% (Robaei et al. 2006). Residual amblyopia (>0.3 logMAR) was found in 1.1% of a screened population of 12- to 13-year-old children in Sweden (Ohlsson et al. 2001). Residual amblyopia in our study included both cases of

unsuccessfully treated combined-mechanism amblyopia and missed (and thus untreated) cases of refractive amblyopia. In general, children with combined-mechanism amblyopia may have a worse prognosis, even despite treatment.

Refractive amblyopia and combined-mechanism amblyopia occurred more frequently than strabismic amblyopia in our study. The distribution of amblyopia types differs from that in other studies, in which combined-mechanism amblyopia was present in 19% of amblyopic children only (Robaei et al. 2006) or refractive amblyopia accounted for 78% (Multi-ethnic Pediatric Eye Disease Study Group 2008).

In our study, children with strabismic or combined amblyopia were 2 years younger than children with refractive amblyopia when they first visited the orthoptist or ophthalmologist (about the age of 2.5 years and 4.5 years, respectively). In the late 1960s, the various types of amblyopia were detected more than 2 years later, although with the same age sequence (Simonsz-Toth et al. 2007). In this historic cohort study, occlusion therapy was started at a mean age of 5.1 years for strabismic amblyopia, 5.7 years for combined-mechanism amblyopia, and 6.6 years for anisometropic amblyopia. In a retrospective Australian study of 127 children with amblyopia, a trend was also seen for earlier detection of deprivation amblyopia and later detection of anisometropic amblyopia, although amblyopia type and age at the first outpatient visit were not significantly related (Chua et al. 2004).

We cannot give a definite assessment of how effective the Dutch child vision screening program is when there are regular vision measurements until age 6. In a British study, the prevalence of amblyopia in 7.5-year-old children was significantly smaller in children who underwent intensive orthoptic screening between 8 and 37 months compared with orthoptic screening at 37 months only (Williams et al. 2002). Our results suggest that preschool screening from age 3 contributes most to amblyopia detection. Preverbal screening especially contributed to the earlier detection of strabismic and combined-mechanism amblyopia. Whether earlier detection and treatment of amblyopia is cost-effective, remains to be seen (IQWiG 2008).

Acknowledgments

The authors thank all Rotterdam child health care physicians; the orthoptists and ophthalmologists at the Rotterdam Eye Hospital, Erasmus MC, Ikazia Ziekenhuis, Medisch Centrum Rijnmond Zuid locatie Zuider, Medisch Centrum Rijnmond Zuid locatie Clara, St. Franciscus Gasthuis, Ruwaard van Putten Ziekenhuis and IJsselland ziekenhuis and the research orthoptists Ellen van Minderhout, Brigitte Simonsz, Jan-Roelof Polling, Marleen Vermeulen, and Isa Vels en Jolanda de Vos-van den Broek for their efforts in collecting the

data; and the research assistants Ilse Oonk, Priscilla Dorresteijn and Mirjam Goudswaard for their assistance with data collection and the organization of the study.

Supported by Grant ZonMw from The Netherlands Organization for Health Research and Development (Project 28.2689.0).

Chapter 3

Determinants and outcome of unsuccessful referral after positive screening in a large birth-cohort study of population-based vision screening

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J AAPOS. 2011;15(3):256-262

ABSTRACT

Purpose. The efficacy of population-based vision screening is hampered by unsuccessful referral after a positive screening test. We studied the nature and causes of unsuccessful referral in a 7-year birth cohort study of vision screening in Rotterdam, The Netherlands.

Methods. All parents of children who had been unsuccessfully referred were asked whether they recalled the referral. Reasons for noncompliance, if any, were identified using semi-structured interviews. Screening records were checked for written evidence of the referral. The parents' fluency in Dutch and their socioeconomic status were also assessed.

Results. Of the 561 screen-positive children, 129 (23%) had not been referred successfully. For the current study, 97 parents were successfully contacted. Of these, 14 parents had been willingly noncompliant and 83 said they were unaware of the referral, with 47% having poor to moderate fluency in Dutch. In 53 cases, the screening charts contained no written evidence of any referral. Amblyopia was identified in 3 of the unsuccessful referrals.

Conclusion. In this population-based screening program, 1 of 4 positively screened children was not successfully referred. Apart from parental noncompliance, the unsuccessful referrals can be explained by miscommunication, deficient documentation, and physician noncompliance with screening guidelines. An effective monitoring feedback system may improve the efficacy of child vision screening.

INTRODUCTION

Amblyopia is commonly defined as a persistent, unilateral, or bilateral reduction in vision after the correction of any refractive error caused by an interruption in the normal visual development during the first years of life and is not caused by an organic ocular abnormality (Holmes et al. 2006a). Patients with untreated amblyopia generally experience a decrease in quality of life (Van de Graaf et al. 2007) and are at risk for bilateral visual impairment if visual function in the better eye is comprised (Holmes et al. 2006a; van Leeuwen et al. 2007). To improve detection of amblyopia, regular childhood vision screening for amblyopia was implemented in The Netherlands through the Dutch child health screening program (Appendix II) (Van Velzen-Mol 2002; Van Velzen-Mol et al. 2006). Currently, most Dutch children with amblyopia are now diagnosed at a treatable age (see Chapter 2 of this thesis). Nevertheless, approximately one third of these children do not acquire a visual acuity of 0.3 logMAR at adult age either because of faulty screening or due to noncompliance with amblyopia treatment (Lithander et al. 1991; Loudon et al. 2006; Stewart et al. 2005; Vinding et al. 1991; Woodruff 1995). Loudon and colleagues (2006) showed that low compliance with occlusion therapy for amblyopia in children is strongly correlated with poor parental fluency in the national language, low levels of education, and the initial visual acuity. Compliance with a referral after a positive screening test, however, may influence an amblyopia outcome as well (Juttmann 2001; Mark et al. 1999; Snowdon et al. 1997a).

Williamson and colleagues (Williamson et al. 1995) showed that high rates of default in terms of vision screening appointments, particularly in geographical areas of lower socioeconomic classification, have hampered the attempts at reducing the incidence of insufficiently treated amblyopia. Early results of a large birth-cohort study on vision screening showed that approximately one third of the children had not been successfully referred after a positive screening test (Juttmann 2001). In the current study, we investigated noncompliance after a positive vision-screening examination to determine reasons for unsuccessful referral.

METHODS

We investigated the unsuccessful referrals of children from the Rotterdam AMblyopia Screening Effectiveness Study (RAMSES) (Juttmann 2001), a 7-year follow-up study of a cohort of 4,624 children born in Rotterdam between 16 September 1996 and 15 May 1997. That study had been set up to determine the sensitivity, specificity and effectiveness of the Dutch population-based child vision-screening program (Appendix II). For the cur-

Table 3.1) Five-point scale to measure fluency in the Dutch language (Loudon et al. 2006).

Score	Description	Definition
1	None	Conversation with a parent could only be accomplished through an interpreter
2	Poor	Parent gave only superficial responses to the questions, using same standard phrases and sentences
3	Moderate	Parent unable to formulate proper Dutch sentences
4	Good	Non-native parent spoke fluent Dutch, but obviously not as his/her native language
5	Excellent	Native speaker of Dutch

rent study, we included children from the RAMSES birth cohort who had had a positive vision-screening test but who had not been successfully referred to a practitioner, an ophthalmologist, or an orthoptist and who had undergone the final orthoptic examination of the RAMSES at age 7. Parents of these unsuccessfully referred children were contacted by phone (maximum of 3 attempts) or home visits (maximum of 2 attempts). Parents were asked about their child's vision-screening history, on whether their child had ever been referred after a positive vision screening, and whether they had complied with this referral. If parents could not be contacted after several attempts, we used the data of the RAMSES additional survey that was sent to the parents when the child was six years old. The leading questions in that survey were similar to the leading questions that had been asked by telephone or during the home visits in the current study: "Do you remember any positive screening test?", "Do you remember ever being referred?", and, if these were answered affirmatively: "What did you do after being referred?". The current study, which began after the conclusion of RAMSES, was designed in such a way so as not to interfere with the actual practice of vision screening, referral and follow-up.

Previous studies (Loudon et al. 2006; Williamson et al. 1995) have demonstrated that compliance is highly correlated with parental fluency in the national language and socioeconomic status. In our study, parental fluency in Dutch was rated by the researchers on a five-point scale (Table 3.1) (Loudon et al. 2006). If fluency in Dutch was deemed to be "poor" to "none", we attempted to arrange a home visit to ask these questions in person and asked parents to have a relative or neighbour to translate and interpret our questions (eventually, the questions were interpreted for the parent by a third person in 15 cases). Socioeconomic status was determined according to the classification of the residential areas, which was based on residential zip codes with various districts of Rotterdam classified as being either "affluent", "moderate" or "deprived", according to mean house value, mean income, percentage of unemployment, and safety index per district (I*RIS Rotterdam The Netherlands 2009).

To determine the impact of unsuccessful referral on the effectiveness of screening, we assessed what proportion of insufficiently treated amblyopia (i.e. >0.3 logMAR) at age 7 had resulted from unsuccessful referral. Cases of amblyopia had been identified by

using the diagnostic information provided by treating orthoptists and/or the vision data collected in the final study examination at age 7. The cut-off value of 0.3 logMAR is in line with the cut-off used in the studies by Rahi and colleagues (Rahi et al. 2002a) and Van Leeuwen and colleagues (van Leeuwen et al. 2007) and in line with the visual-field restriction precluding driving and conformed with the standards in The Netherlands and the United States.

Based on the parents' answers to the questionnaire regarding their child's vision-screening history, cases were divided into two groups (Fig. 3.1): "*Unaware of Referral*" and "*Willingly not Compliant*".

We considered cases to be "*unaware of referral*" when parents indicated not having remembered any referral or positive screening test. In those cases, the children's screening

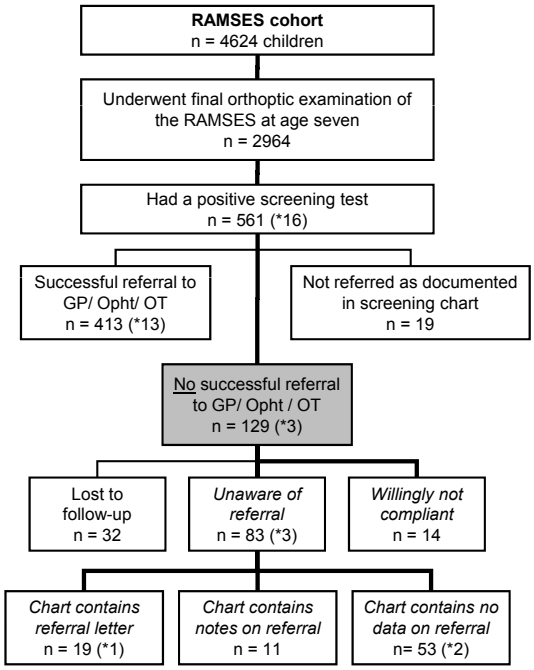

Figure 3.1) Flowchart illustrating the study design. Children who underwent the final orthoptic examination of the RAMSES birth-cohort study at age 7 and who were positively screened were subdivided according to whether they visited a general practitioner, ophthalmologist, or orthoptist and whether they had been successfully or unsuccessfully referred after the positive screening test. The gray box represents the 129 children who were included in the current study. These were classified according to their parents' awareness of the referral recommendation and documentation in the screening charts of referral. GP, general practitioner; Opht, ophthalmologist; OT, orthoptist. * Number of cases of insufficiently treated amblyopia having an acuity.>0.3 logMAR at age 7.

records were checked to verify whether any referral recommendation had been given on the same date as the positive screening test. Based on the documentation in the screening records and the presence of a copy of the referral letter, these cases were further subdivided into three categories:

(1) "*Chart contains referral letter*" included children who had been referred with great certainty, as a copy of the referral letter was present in the screening records;

(2) "*Chart contains notes on referral*" included children for whom referral was uncertain since only the referral recommendation, and not a copy of the referral letter, was documented in the screening records.

(3) "*Chart contains no data on referral*" included children for whom referral was even more uncertain because the screening records contained no written evidence of the positive screening test and/or referral

We considered cases to be "*willingly not compliant*" when parents admitted to not complying with the referral after the positive screening test. Information on the parents' levels of education and countries of origin was requested during the phone or house visits. These parents were asked to participate in a semi-structured, qualitative at-home interview to explore why they had not complied with referral. The semi-structured interview covered topics in three primary domains identified from an earlier questionnaire (Loudon et al. 2009): course of events during the positive screening test (domain: "*child*"), knowledge and perception of the parent about the referral (domain: "*parent*"), and communication with the physician (domain: "*physician*"). The interviews were analyzed based on the principles of grounded theory (Holloway 2005), which generated a systematic list of all the possible reasons for being noncompliant.

RESULTS

The eligible participants included 129 positively screened children who had not been successfully referred to the general practitioner, ophthalmologist, or orthoptist and who had undergone the final orthoptic examination during RAMSES at age 7.

Of the 4,624 children from the RAMSES birth cohort, 2,964 underwent the final orthoptic examination at age 7 (Fig. 3.1). Of the 2,964 children, 561 had had a positive screening test, of whom 129 (23.0%) had not been successfully referred. Parents could be contacted by phone or home visit in 64 of 129 cases, whereas only data from the parental survey from the RAMSES study were available in 33 cases. The remaining 32 patients could not

be reached, and no parental survey was available. The visual acuity outcome at age 7 of these 32 missing patients was <0.3 logMAR in all cases.

Of the 97 parents for whom referral follow-up data were available, 83 parents were *unaware of referral* and 14 were *willingly not compliant* (Fig.3.1). Of the 97 children, 34% lived in low-socioeconomic status areas, 41.2% in "moderate" areas, and 24.7% in "affluent" areas. Parental fluency in Dutch was rated to be "none/poor" to "moderate" in 24 of 51 parents (47%) who were *unaware of referral*, and in 3 of 13 parents (23%) who were *willingly noncompliant*. In our study, the difference in socioeconomic status between unsuccessfully and successfully referred children nearly reached the threshold of statistical significance (P = 0.097, X^2 test).

Figure 3.2 shows the relationship between visual acuity of the poorest eye at the time of screening and visual acuity of the poorest eye at age 7. In children younger than 3 years of age, visual acuity measurements were not performed at screening (pertaining to 30 of the 97 unsuccessfully referred children), whereas visual acuity measurements were unknown in 14 more children who had had their first positive test at preschool screening.

Unaware of Referral

The parents of 83 children said that they were not aware of having been referred. The majority of these children lived in areas of "deprived" to "moderate" socioeconomic status, and their parents had a fluency in Dutch classified as moderate or less (Fig. 3.3A-C). In 19 of these 83 cases, a copy of the referral letter was found in the screening records. One child had a visual acuity of >0.3 logMAR at age 7. This amblyopic child lived in a deprived residential area and its parents had poor parental fluency in Dutch. In 11 cases the referral recommendation had been documented in the screening records, but there was no copy of the referral letter. None had a visual acuity of >0.3 logMAR at age 7. In the remaining 53 cases, no documentation about the referral recommendation (and, in many cases, no documentation about the positive screening test) was found in the screening records. In this group, 2 children had a visual acuity of >0.3 logMAR at age 7.

Willingly Not Compliant

Fourteen cases were found to be willingly not compliant with referral. Of these, 11 lived in areas of "moderate" to "affluent" socioeconomic status. Fluency in Dutch could be rated in 13 of the 14 cases: 10 had good to excellent fluency in Dutch (Fig. 3.3D). Ten had a high level of education. Six parents belonged to an ethnic minority group. No child in this category had a visual acuity of >0.3 logMAR at age 7. One child, however, did have amblyopia, but the visual acuity in the poorest eye was <0.3 logMAR at age 7. Of the 14 parents, 3 refused to participate in the semi-structured interview about compliance

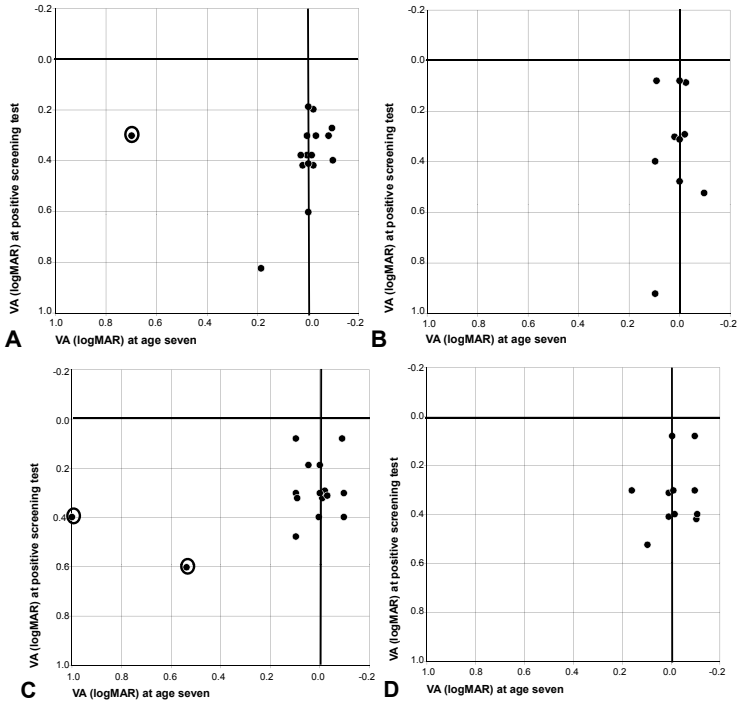

Figure 3.2) Relationship between visual acuity of the poorest eye at the time of screening and at age 7 (data on visual acuity at the first positive screening were not available for 44 children). Each dot represents a child: cases with amblyopia who had a visual acuity >0.3 logMAR at age 7 are circled. A, Unaware of referral (chart contains letter). B, Unaware of referral (chart contains referral notes). C, Unaware of referral (chart contains no referral data). D, Willingly not compliant.

with referral. From the interviews with the remaining 11 parents, 3 of the most frequently mentioned reasons for noncompliance were lack of trust in the screening test or the test results, lack of knowledge of the illness or its treatment, and fear of diagnosis or treatment (Table 3.2).

Last, mean visual acuity of the poorest eye at the time of screening was 0.49 SD, 0.59 logMAR in those children who had been referred successfully (Fig. 3.4), and 0.32 SD, 0.77 logMAR in those children who had not been referred successfully (Fig. 3.4). This difference was significant ($P < 0.0001$, unpaired *T*-test).

Of the 2,964 children who underwent the final study examination at age 7, amblyopia was diagnosed in 100 (Chapter 2). Of these, 73 children had been screened positively at least once; 16 of these had insufficiently treated residual amblyopia at age 7 (visual acuity of the poorest eye .>0.3 logMAR), including 3 cases of unsuccessful referral (Fig. 3.1).

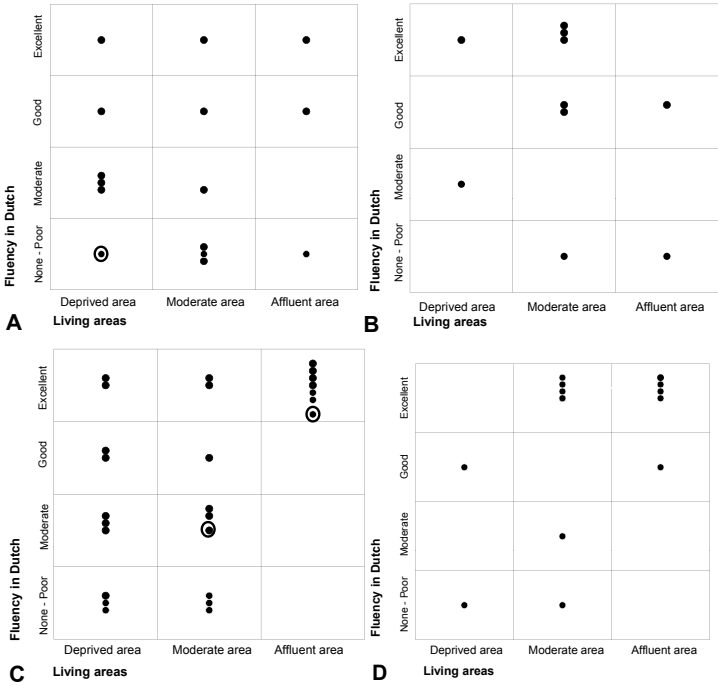

Figure 3.3) Parental fluency in Dutch versus SES living area (data on parental fluency not available for 33 cases). Each dot represents a child. A, Unaware of referral (chart contains letter). B, Unaware of referral (chart contains referral notes). C, Unaware of referral (chart contains no referral data). D, Willingly not compliant.

Table 3.2) Reasons for noncompliance with referral after a positive vision-screening test given by 11 parents who were willingly not compliant (qualitative interviews)

Reasons for being noncompliant with referral	Frequency
Lack of trust in screening test or test result	7
Lack of insight into treatment or illness	5
Fear of diagnosis or treatment	3
Lack of trust in regular health care	1
Preference for alternative medical treatment	1
Financial problems	1
Forgot	1
Measurement of visual acuity by parent at home	1
Difficulty with arranging an appointment	1
Dissatisfied with behaviour of screening physician	1

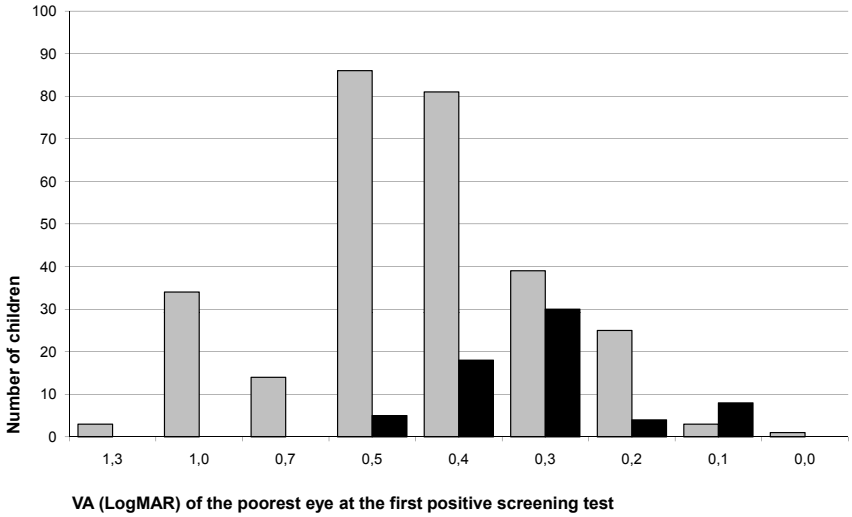

Figure 3.4) Comparison between the visual acuity of the poorest eye at the time of positive screening of children who had not been successfully referred after the positive screening test (black bars, n = 65 of the 129 children in whom visual acuity had been measured), and children who had been referred successfully after the positive screening test (gray bars, n = 286 of the 413 children in whom visual acuity had been measured).

DISCUSSION

Willing noncompliance, although possibly underreported, was relatively rare in our study, representing only one tenth of unsuccessful referrals. These parents had been aware of the referral recommendation by the screening physician but very often did not agree with the screening results. Most of them lacked sufficient insight into amblyopia, its treatment, and the importance of screening.

In most of the cases of unsuccessful referral, the parents stated that they were unaware of the referral. This lack of awareness seemed plausible in cases where the children's screening records did not contain written evidence of any referral, even though a positive screening test had been reported to the study centre. It is possible that the screening physician deviated from the screening protocol in these cases, especially when visual acuity at screening was near the referral threshold.

In other instances, however, the child's screening record did contain written evidence of a referral. Although a note on the referral or a copy of any referral letter does not guarantee that the referral actually took place, it is possible that neither parent understood the referral recommendation. In this group, one half of the parents had poor to moderate fluency in Dutch and lived in areas of low socioeconomic status. However, approximately

47% of the inhabitants of Rotterdam are non-native (I*RIS Rotterdam The Netherlands 2009). Differences in socioeconomic status, between the successfully and unsuccessfully referred children, approached statistical significance in our study. Other studies are more conclusive in this regard, finding that socioeconomic classification and fluency in the national language do affect compliance (Loudon et al. 2006; Mark et al. 1999; Subramanian et al. 2004; Sutton et al. 1994; Vernon et al. 1990; Williamson et al. 1995). Parents of non-Western children are less likely to consult an ophthalmologist than parents of Dutch children (Van Laar 2007). With this in mind, the proportion of unsuccessful referral may well be lower in other non-cosmopolitan areas in The Netherlands.

An alternative explanation for the parental unawareness is that some of the parents may not have been fully truthful about not understanding the referral recommendation for their child. Finally, recall bias may have played a role.

As mentioned above, it is possible that some screening physicians had not referred a child if the visual acuity was near the referral threshold. In Sweden, the referral threshold was increased in 1992 from visual acuity 0.1 to 0.2 logMAR for a 4-year-old to reduce the number of over-referrals (Hard 2007; Hard et al. 2002). The current threshold for referrals in The Netherlands is a visual acuity of 0.2 logMAR in 1 eye or a visual acuity of 2 lines difference between the eyes (Appendix I) (Van Velzen-Mol 2002). In most cases, visual acuity will improve slightly from the end of treatment until adulthood, but in rare cases visual acuity may decline (Simonsz-Toth et al. 2007). With these rare cases in mind, the threshold for referral at child vision screening has been set to low levels.

The screening physicians did not refer 19 other children with a positive preschool screening test (3.3% of 561), as had been explicitly stated in the screening record (Fig. 3.1). In these cases, the physicians had deviated from the guidelines for referral after a positive screening test and had, instead, decided to re-examine the child within 3 months. Thirteen of these children lived in areas of low socioeconomic status. Testing had failed due to lack of cooperation by the child in 12 cases. In such cases, physicians may decide not to comply with the screening protocol, especially when the parents and the child do not speak Dutch or when the visual acuity was near the threshold for referral.

It is apparent that noncompliance within screening programs occurs at the level of both screening physicians and parents (Subramanian et al. 2004). In conclusion, a decision made by a physician or parent not to act upon a positive vision screening test may have been guided by a normal (negative) screening history, normal subsequent screening examinations, or the absence of clinical symptoms of visual impairment. Parents may have had the visual acuity of their child reassessed locally. Moreover, many of the unsuccess-

fully referred children had a minimal decrease of visual acuity at screening, and screening staff may have felt that referral in these cases was not necessary since the chance of amblyopia was deemed absent.

Our study has several limitations. RAMSES was a very large cohort study with a 7-year follow-up. Its main goal was to study the effectiveness of child vision screening in The Netherlands without any interference in routine screening practice and follow-up. The nature and cause of unsuccessful referrals, therefore, only could be studied after the termination of the RAMSES study. It must be realized that at the start of the birth-cohort study in 1995, it was not anticipated that more than 10% of referrals would be unsuccessful. As the main study did not address ethnic or socioeconomic inequalities, data on the socioeconomic status had to be collected retroactively, as did data on the country of origin and parental fluency in Dutch.

The impact of unsuccessful referral in terms of visual acuity outcome was small, although not negligible. About 1 of 5 cases (3/16) of insufficiently treated amblyopia in the birth cohort could have been prevented if compliance with the guidelines for referral had been good. Therefore, referral after a positive screening test should be documented more thoroughly and requires an effective monitoring feedback system. These may prevent the deprivation of proper care to children and provide feedback on their decisions to physicians.

Acknowledgments

The authors thank Hazebroek-Kampschreur, Irene Tjiam, and Luc Vercauteren for their advice, and the screening nurses of the public health services in Rotterdam for their help in verifying the screening charts. Acknowledgement for proofreading and correcting the English edition goes to Mrs. K. Verhoef of VERT-ICAL Translations & Administration.

Chapter 4

How Dutch orthoptists deal with noncompliance with occlusion therapy for amblyopia

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Strabismus. 2010; 18(4), 146–166

ABSTRACT

Background. We previously found that compliance with occlusion therapy for amblyopia is poor, especially among children of non-native parents who spoke Dutch poorly and who were low educated. We investigated conception, awareness, attitude, and actions to deal with noncompliance among Dutch orthoptists.

Methods. Orthoptists working in non-native, low socioeconomic status (SES) areas and a selection of orthoptists working elsewhere in The Netherlands were studied. They were observed in their practice, received a structured questionnaire, and underwent a semi-structured interview. Finally, a short survey was sent to all working orthoptists in The Netherlands.

Results. Nine orthoptists working in non-native, low-SES areas and 23 working elsewhere in The Netherlands participated. One hundred and fifty-one orthoptists returned the short survey. Major discrepancies existed in conception, awareness, and attitude. Opinions differed on what should be defined as noncompliance and on what causes noncompliance. Some orthoptists found noncompliance annoying, unpleasant, and hard to imagine, others were more understanding. Many pitied the noncompliant child. Almost all thought that the success of occlusion therapy lies both with the parents and the orthoptist, but one third thought that noncompliance was not solely their responsibility. Patients' compliance was estimated at 69.3% in non-native, low-SES areas (electronically, 52% had been measured), at 74.1% by the other 23 orthoptists, and at 73.8% in the short survey. Actions to improve compliance were diverse; some increased occlusion hours whereas others decreased them. In non-native, low-SES areas, 22% spoke Dutch moderately to none; the allotted time for a patient's first visit was 21'; the time spent on explaining to the parents was 2'30" and to the child 10". In practices of the other 23 orthoptists, 6% spoke Dutch moderately to none ($P < 0.0001$), the time for a patient's first visit was 27'24" ($P = 0.47$), and the periods spent explaining were 2'51" ($P = 0.59$) and 26" ($P = 0.17$), respectively.

Conclusion. Conception, awareness, attitude, and actions to deal with noncompliance varied among orthoptists. In non-native, low-SES areas, time spent on explanation was shorter, despite a lower fluency in Dutch among the parents.

INTRODUCTION

We previously found that electronically measured compliance with occlusion therapy for amblyopia is poor: children were patched on average 57% of the occlusion time prescribed by the orthoptist (Loudon et al. 2006). 15.7% of the children were patched less than 30% of the time that was prescribed (Loudon et al. 2006). Low compliance correlated highly with low initial visual acuity of the child, poor parental fluency in the Dutch language, parental country of origin, low SES, a low level of education of the parent, and psychological distress in the family caused by occlusion therapy (Loudon et al. 2006; Loudon et al. 2009; Smith et al. 1995). In this study, we investigated how orthoptists deal with low compliance.

Healthcare professionals rarely underestimate compliance in compliant patients (Kass et al. 1986a) but frequently overestimate compliance in noncompliant patients (Copher et al. 2010; Kass et al. 1986a; Moreau et al. 2009; Murri et al. 2002; Wens et al. 2005; Wright 1993; Wylie et al. 2002). Identifying patients with poor compliance is a difficult task for the healthcare professional (Copher et al. 2010; Kass et al. 1986a; Urquhart 1997). Most healthcare professionals are guided by the results of the therapy to identify defaulting patients, and the professional's instinctive feeling about the patient's compliance plays an important role in its prediction (Gelb et al. 2008; Kass et al. 1986a; Wens et al. 2005). Some healthcare professionals ignore noncompliant behaviour completely (Gelb et al. 2008; Heszen-Klemens 1987; Wright 1993), as noncompliant behaviour contravenes professional beliefs, norms, and expectations regarding the "proper" roles of patients and professionals (Playle et al. 1998; Stimson 1974; Wens et al. 2005). A study on compliance with glaucoma treatment identified three types of physicians: physicians who were less proactive about compliance, sceptical physicians who were less likely to believe that compliance could be changed, and idealistic physicians who addressed compliance across their patient population (Gelb et al. 2008).

How noncompliance with occlusion therapy for amblyopia manifests itself in children or their parents has been part of several studies (Awan et al. 2005; Awan et al. 2010; Dixon-Woods et al. 2006; Dorey et al. 2001; El-Ghrably et al. 2007; Karlica et al. 2009; Loudon et al. 2006; Loudon et al. 2009; Stewart et al. 2005; Stewart et al. 2004b; Stewart et al. 2007a), but not how orthoptists deal with it. Hence, we made an inventory, among Dutch orthoptists, of their conception, awareness, attitudes, and actions to deal with noncompliance. As noncompliance is correlated with country of origin, parental fluency in Dutch, and level of education, we were especially interested in orthoptists working in non-native, low socioeconomic status (SES) areas and compared them with orthoptists working elsewhere in The Netherlands.

METHODS

In this observational study, we recruited two groups of orthoptists and made assessments with a semi-structured interview, a structured non-validated questionnaire, and observations in their orthoptic practice. Data were compared between these two groups and with those obtained with a short survey sent to all working orthoptists in The Netherlands.

Participating Orthoptists

Two groups of Dutch orthoptists were recruited to the study: orthoptists working in clinics located in non-native, low-SES areas of Amsterdam, Rotterdam, The Hague, and Utrecht, and orthoptists working in clinics located in other areas of The Netherlands. For the first group, we selected clinics located in the four large cities of The Netherlands, in districts with a large proportion of non-native and low-SES inhabitants (Statistics Netherlands 2005). Orthoptists working in these clinics were asked to participate in the study. For the second group, we recruited orthoptists working elsewhere in The Netherlands by making an appeal during a meeting of the *Nederlandse Vereniging van Orthoptisten*, the National Orthoptic Association of The Netherlands (NVvO). Finally, a short survey was sent to all orthoptists who were members of the NVvO.

Observations in the Orthoptic Practice

The observations of the orthoptists in practice were (1) non-participating: i.e., during the observations the researcher did not interact between the orthoptist and the patient; (2) non-open: i.e., the orthoptist did not know what items were observed; (3) partly structured: i.e., the observer recorded specific events; and (4) partly non-structured: i.e., the observer noted purposive, descriptive observations. Structured observations were made regarding age and gender of the patient, fluency in the Dutch language of his/her parents, the purpose of the visit, the allotted time per visit, the time actually spent per visit, and the time spent for explanation of the diagnosis and treatment to the child and parent. Descriptive non-structured observations were made of any explanation given regarding diagnosis and treatment to the parent and to the child. Each orthoptist was observed in his or her practice for one day.

Semi-structured Interview

The purpose of the face-to-face, semi-structured interview was to gain qualitative information about: structure of the clinic (e.g., allotted time per new and follow-up visit), cooperation within the clinic (e.g., between orthoptists and ophthalmologists or assistants), demographic characteristics of their patients (e.g., the proportion of non-native patients), attitude towards parents having a poor fluency in Dutch or a low SES, and conception, attitude, and approach towards noncompliance. This semi-structured interview was de-

veloped in a focus group (co-authors AMT, EV, MMS, WLA-T, and HJS). Two researchers (WLA-T and AMT) independently analyzed the interviews according to the principles of Grounded Theory (Holloway 2005). All responses of the orthoptists to each question were coded systematically and ordered in whether the response concerned a "fact," an "opinion," or an "action." This generated a systematic list of all possible statements per subject and how frequent these statements were reported by the orthoptists. The results of the analyses of both researchers were compared and discrepancies were supplemented and resolved.

Structured Questionnaire

This questionnaire was developed in a focus group (MMS, HJS, and AMT). The first part of the structured questionnaire ("Orthoptists' individual personality features") contained questions to identify the personality of the orthoptist, and was subcategorized as: "autonomy," "being well organized," "ability to comprehend," "flexibility," "early adopter," "hierarchy," and "assertiveness." This part was developed based on a questionnaire of the Dutch Institute of Care and Welfare (Verbeek 1999) that assessed the quality of healthcare practices in general. The second part ("attitude towards noncompliance") of the questionnaire consisted of statements that described different practical situations regarding noncompliance with occlusion therapy. The first part of the questionnaire was based on categories that were selected from general assessments regarding professional attitudes of healthcare givers. The statements of the second part of the questionnaire were developed based on the Stages of Change model of Prochaska & Velicer (Prochaska et al. 1997). The questionnaire contained 125 questions that were scored on a 5- or 6-point scale, i.e., "strongly agree" to "strongly disagree," and "(almost) always" to "(almost) never," respectively.

Short Survey

Almost all orthoptists in The Netherlands, approximately 95%, are members of the NVvO. All members of the NVvO (n = 326) received a short, structured questionnaire survey that contained 37 questions. The questionnaire was returned and processed anonymously, although later several questionnaires returned by the same orthoptists could be recombined by identification of a self-generated code. The questionnaire assessed general information of the orthoptist, orthoptists' conception of compliance and noncompliance, orthoptists' estimation of noncompliance, the proportion of non-native patients in their practice, actions to deal with noncompliance, the attitude towards noncompliance, and relationship between patient and orthoptist. It was developed in a focus group (WLA-T, MMS, HJS, and AMT).

Main Outcome Measures

Primary outcome measures of the study were conception, awareness, attitudes, and actions to deal with noncompliance. Secondary outcome measures were fluency in Dutch of the parents of the patients, the allotted time per visit, the time actually spent per visit, and the time spent explaining to the child and to the parent the diagnosis and therapy.

Statistical Analysis

Data of the questionnaires and the structured observations were entered into SPSS 16.0 for Windows. Non-parametric statistics (X^2 test and *Kruskall-Wallis* test) were used to demonstrate differences between the groups of orthoptists in answers to the structured questionnaires and between patients' characteristics that had been observed during the observations in practice. In the analysis of the questionnaire and in the short survey, the outcome of the questionnaires was used as the dependent variable and the groups of orthoptists as the independent variable. We considered a *P*-value of 0.01 to be statistically significant to take multiple testing into account.

Covariance analyses were used to compare, between groups, the allotted time per visit, the time actually spent per visit, and the time spent on explaining diagnosis and treatment to the child and to the parent. $P < 0.05$ indicated statistical significance. The questionnaire and the short survey were not validated.

As part of another study (Vukovic et al. 2008), compliance was measured electronically with the Occlusion Dose Monitor in the children in non-native, low-SES areas. The electronically measured compliance was defined as the actual occlusion time measured with the Occlusion Dose Monitor divided by the prescribed occlusion time and expressed as a percentage.

RESULTS

Nine orthoptists working in five clinics located in non-native, low-SES areas of the four large cities of The Netherlands (Appendix IV), and 23 orthoptists working in 15 clinics dispersed over The Netherlands (Appendix IV), were recruited for the study. The orthoptists of one clinic in the south of Rotterdam declined to participate and instead two clinics in the vicinity participated. The 23 orthoptists dispersed of The Netherlands volunteered for participating in the study. From all the members of the NVvO, 157 orthoptists returned the short survey, including the 32 study orthoptists. Six of these were excluded from analysis, as they had retired or did not treat amblyopic children. In total, 151 questionnaires from 326 orthoptists (46.3%) were included in the analysis.

Orthoptists' Personalities

No differences in personalities between orthoptists from non-native, low-SES areas and those working elsewhere in The Netherlands could be found (Appendix V). Orthoptists reported autonomy at their work, found themselves well organized, and said they were able to comprehend patients in general, including those from other cultures. They considered themselves assertive and flexible, but indicated that they lacked the time to act flexible at work. In both groups, orthoptists varied in ways of adopting new innovations. Most orthoptists read their professional literature often (25/32). Only a few considered themselves open for change (7/32). Esteem for hierarchy was low. However, the majority agreed that patients should obey the prescription of the orthoptist (22/32).

Conception of Compliance

Both in the interview and in the questionnaire, we found that orthoptists differed widely in what they considered to be noncompliance. Two orthoptists found their patients to be noncompliant if they patched less than 85% of the prescribed time, ten other orthoptists less than 50%, and two less than 30% of the prescribed time. However, 13 orthoptists only considered a patient noncompliant if he or she did not comply with the treatment at all. According to the orthoptists, noncompliance includes failure to show up for an appointment without notice. Orthoptists mentioned many different reasons and circumstances that could cause noncompliant behaviour (Table 4.1): parents with low SES (n = 9); parents with poor fluency in the Dutch language (n = 7); parents with long working hours (n = 6); children being patched at home instead of at school (n = 3); chaotic and inconsistent parents (n = 3); children with low initial visual acuity (n = 3); parents objecting to treatment (n = 3); non-native girls (n = 2); children with mental retardation (n = 2); older children (n = 1); and non-native parents (n = 1). Only six orthoptists held the opinion that noncompliant behaviour exists in all social classes of the community.

Awareness of and Attitude Towards Noncompliance

We found little indication that orthoptists working in non-native, low-SES areas differed from those working elsewhere in The Netherlands. The second group more often believed that their patients understood their explanation (15/23 vs. 1/9, P = 0.028) and that their patients occluded almost precisely as they prescribed (17/23 vs. 2/9, P = 0.014), but most parents of the children treated by the second group of orthoptists spoke Dutch well. Twenty-four of 32 orthoptists declared they could recognize noncompliance. Orthoptists in non-native, low-SES areas stated they had a lot of poor compliance in their practice and that their patients did not attend an appointment often (P = 0.010 and P = 0.018). The estimation of their patients' compliance was 69.3% SD 8.3. It was estimated at 74.1% SD 18.8 by the 23 orthoptists working elsewhere in The Netherlands. It was estimated at 73.8% SD 16.0 by the 151 orthoptists who participated in the short survey (Appendix

Table 4.1) Reasons for noncompliance according to the 32 orthoptists in both groups (derived from the semi-structured interview on compliance with occlusion therapy).

Reasons of noncompliance	Frequency
Circumstances at home: occlusion therapy does not fit into daily life	16
Child does not accept its parent's authority	15
Parents do not understand amblyopia and the purpose of occlusion	7
Forgot	4
Holiday period	4
Parents do not wish to patch	5
Low initial visual acuity	2
Long duration of the therapy	2
Irritation to the skin	2
Child is falling behind at school due to the occlusion therapy	2
Other health-related or behavioural problems in the child	2
Miscommunication between orthoptist and parent	1
Financial problems	1

VI). Compliance was measured electronically in 79 of the patients in non-native, low-SES areas with the Occlusion Dose Monitor and averaged 52% with a bimodal distribution and about a fifth zero compliance (Vukovic et al. 2008).

Almost all orthoptists were of the opinion that compliance is an important issue within the orthoptic practice (30/32). In their opinion, the responsibility for the success of occlusion therapy lies with both the parent and the orthoptist (30/32). However, 10 of the 32 orthoptists were of the opinion that it is not their sole responsibility if the child was not patched as prescribed. Nine of the 32 orthoptists believed that compliance could not be much improved. Nonetheless, 23 of the 32 orthoptists said they wanted to learn about methods to improve compliance and how to deal with noncompliance.

Both in the interview and the short survey, orthoptists found noncompliant behaviour annoying, unpleasant, and hard to understand. Two orthoptists were shocked when their patients were not patched as prescribed. Two other orthoptists were understanding and said they had sympathy for noncompliant patients. Overall, orthoptists pitied the child when noncompliance occurred. However, orthoptists were mostly satisfied when visual acuity in the amblyopic eye increased, regardless of whether a patient was compliant or not.

Actions to Detect Noncompliance

During the interview, orthoptists gave various methods to suspect and detect noncompliance with occlusion therapy: if the visual acuity of the amblyopic eye does not increase (n = 8); if new occlusion patches are not needed (n = 3); by asking the child instead of

the parent how the patching went (n = 15); by additional interrogation of the parent (n = 10); and by asking the parent whether occlusion therapy was successfully carried out for at least 1/4 of the prescribed time (n = 2). One orthoptist did not ask whether patching had succeeded. Interestingly, during the observations of the orthoptists in practice, it almost never occurred that the orthoptists assumed that patching had not succeeded. The methods that had been mentioned during the interviews were hardly ever used during the observations. Orthoptists sometimes mentioned to the researcher that the parents had tried as hard as they could. After observing each orthoptist for a day, with an average of 15 patients per orthoptist, only a quarter of all orthoptists had suspected noncompliance in one patient during that day, the others not in a single patient.

Actions to Deal with Noncompliance

Both in the interview and in the short survey, several actions to deal with noncompliance with occlusion therapy were mentioned by the orthoptists (Table 4.2). However, not all of these actions could be realized under the current working conditions. Some orthoptists wanted to plan a sooner follow-up visit for noncompliant patients, but the waiting time for an appointment was longer than 3 months. The strategy to decrease patching hours in case of noncompliance (to avoid failure due to excessive demands on the family) was adapted by 6 of 9 orthoptists in non-native, low-SES areas, and by 23 of 142 orthoptists who returned the short survey.

Parents who had missed an appointment were contacted for a new appointment in four clinics, but only in exceptional, critical cases. Contacting the parents when an appointment had been missed was not considered, by the orthoptists, as part of their work as an orthoptist. Among those orthoptists who returned the short survey (n = 151), 20.7% said they called parents who missed an appointment, 15.3% sent a letter, 8.0% said the secretary called these patients, and 67.3% of the orthoptists said they did nothing. In the interview and in the short survey it was reported that to purchase material to improve compliance or to call parents who missed appointments is hard to realize because it is too expensive for the department.

Communication with Parent and Child

None of the orthoptists in non-native, low-SES areas involved the child in the explanation of diagnosis and treatment, whereas 10 out of the 23 orthoptists working elsewhere in The Netherlands and 18.9% of the 151 orthoptists who returned the short survey said they did so (P = 0.046).

We measured the time spent on explaining the diagnosis and treatment to the parents and child during the observations in the orthoptic practice, in relation to the parents' ability to

Table 4.2) List of actions undertaken to deal with noncompliance with occlusion therapy by the 32 orthoptists in both groups (derived from the semi-structured interview on compliance with occlusion therapy).

Methods used to deal with noncompliance with occlusion therapy	Frequency
Explain amblyopia and occlusion therapy to the parent once more	24
Identify the reason(s) for noncompliance and find solutions for these	19
For example, determine how occlusion therapy best fits into their daily lives	13
Make the next appointment sooner, or make more frequent appointments	11
Terminate occlusion therapy	11
For example, switch to atropine therapy	10
Explain the consequences of not patching	8
Give the parents informational material about amblyopia and occlusion therapy	7
Confront, alarm or frighten the parent when no patching has taken place	6
Allow the child to choose the colour of the eye patches	6
Distribute a commercial poster on patching designed by a firm that produce eye patches	5
Reward the child with small toys when it has patched well	5
Involve the child's school	5
Emphasize the parents' responsibilities	5
Threaten to stop treatment if the child had not worn the patch	5
Increase the prescribed occlusion hours	5
Pay more attention to the parent	4
Explain the benefits of patching	4
Invent a reward system for the parent to use at home	2
Never occlude > 4 hours a day	2
Decrease the prescribed occlusion hours	1
Start with low number of prescribed occlusion hours	1
Involve the general practitioner	1
Continue occlusion therapy for a few months more before its termination	1
Additional explanation is given and carefully documented for in case of legal claim	1

speak Dutch. In non-native, low-SES areas, 21.6% spoke Dutch moderately to none, the allotted time for a patient's first visit was 21′, the time spent on explaining to the parents was 2′30″ SD 1′04″ and to the child 10″ SD 0′11″ (Table 4.3). In practices of the other 23 orthoptists, 6.2% spoke Dutch moderately to none, the time per first visit was 27′24″, and the periods spent explaining were 2′51″ SD 2′04″ and 26″ SD 0′43, respectively.

The allotted time for a patient's first visit was 21′ SD 9′57″ in non-native, low-SES areas, against 27′24″ SD 6′44″ among the 23 other orthoptists (P = 0.47, Table 4.3). When parents did not speak Dutch, orthoptists preferred to have their explanation translated by a family member, friend, or neighbour. Orthoptists stated in the interview that arranging an interpreter to translate via telephone cost too much time and they made no use of

it. Sometimes it took 20 minutes before the correct translator was available. During the observations in practice, it was noted on several occasions that orthoptists gave their usual explanation in Dutch although the parent spoke Dutch moderately or not at all. In cases when parents did not speak the Dutch language well and no translator was available, we observed in practice that orthoptists gave the verbal explanation in Dutch as usual. Pictures, posters, or drawings were rarely used to clarify the explanation.

DISCUSSION

This observational study demonstrated a variety in conception, awareness, and attitude towards noncompliance and actions to deal with noncompliance among Dutch orthoptists. For some orthoptists, less than 85% compliance was considered as noncompliance, whereas others only considered parents and their children as noncompliant when compliance was 0%. Orthoptists found compliance an important issue within the orthoptic practice, but estimated their own patients' compliance high: During the observations it almost never occurred to the orthoptists that their patients had not patched. Only a quarter of all orthoptists suspected noncompliance in a single patient during the day of observation, the others not at all.

Some orthoptists considered noncompliance annoying, unpleasant, and hard to imagine, while others were more understanding. Almost all orthoptists felt that success of occlusion therapy lies with both the parent and the orthoptists; one third thought that noncompliance was not solely their responsibility. Some increased occlusion hours and some decreased occlusion hours in case of noncompliance. We found little difference in personality between the nine orthoptists working in non-native, low-SES areas and the 23 orthoptists working elsewhere in The Netherlands. Differences in responses to questions in the interviews and the questionnaire were primarily caused by the more difficult circumstances that the first group had to work in. In another study (Vukovic et al. 2008), we reported on the specific findings in the children and measured compliance electronically in the children in non-native, low-SES areas. It averaged 52% with a bimodal distribution and about a fifth zero compliance, whereas the orthoptists treating these children estimated it at 69%. This discordance between assumed and actual levels of compliance has been reported in other studies (Copher et al. 2010; Kass et al. 1986a; Moreau et al. 2009; Wylie et al. 2002).

As any other caregiver would do similarly, orthoptists only start to worry about their patients' compliance when the visual acuity in the amblyopic eye does not increase. The increase in visual acuity is influenced, however, by several confounding variables: initial visual acuity, type of amblyopia, type of refraction, and age of the child. Secondly, as

Table 4.3) Results of observations at the practices of participating orthoptists (Groups A and B). The P-value indicates a significant difference in scores between both groups.

	Group A (n = 9)	**Group B** (n = 23)	P-value
Number of patients observed	132	344	
Missed appointment (without prior cancellation)	14.7%	5.3%	**0.016***
Percentage of patients			0.904
≤ 12 yrs	85.3%	85.8%	
> 12 yrs	14.7%	14.2%	
Mean allotted time (minutes) per first visit †	21:00 ± 9:57	27:24 ± 6:44	
Mean allotted time (minutes) per follow-up visit †	15:30 ± 1:35	16:44 ± 2:26	
Mean time spent (minutes) for a first visit †	19:55 ± 9:53	23:07 ± 9:48	0.473
Mean time spent (minutes) for a follow-up visit †	14:49 ± 5:45	15:37 ± 6:13	0.499
Gender †			0.450
Male	48.6%	47.6%	
Female	51.3%	52.4%	
Parental fluency in Dutch †			**<0.0001***
Excellent	51.0%	88.9%	
Good	49.0%	4.9%	
Moderate	19.6%	3.7%	
Poor	0.0%	2.5%	
None	2.0%	0.0%	
Purpose of the visit †			**0.010***
Occlusion therapy	19.7%	15.8%	
Check-up: acuity after end occlusion treatment	0.0%	12.9%	
Check-up: glasses or angle of strabismus	36.3%	31.7%	
Convergence exercises	0.0%	5.0%	
Referral from screening physician or GP	13.6%	5.9%	
Symptoms	16.7%	10.9%	
Refraction in cycloplegia	3.0%	10.9%	
Else	10.6%	7.0%	
Duration of explanation (in minutes) by the orthoptist †‡			
to the parent	2:30 ± 1:04	2:51 ± 2:04	0.59
to the child	0:10 ± 0:11	0:26 ± 0:43	0.17
Manner of explanation *†; by use of			
compulsory formulations §	73.9%	73.7%	0.291
appreciative formulations |	73.9%	74.4%	0.065
Repeated the explanation (once or more) †	34.1%	30.8%	0.646

Tools used to clarify explanation†			0.191
Pictures. posters or figurines	2.3%	2.3%	
Drawings during explanation	1.1%	2.3%	
Translator (family member or friend)	0.0%	1.5%	
Official medical interpreter	0.0%	0.0%	
None	96.6%	93.2%	
Verified whether explanation was understood †			0.064
Yes: "Yes?" or "Okay?"	26.1%	44.4%	
Yes: "Do you understand?" #	4.6%	3.0%	
No	69.3%	52.6%	

* P < 0.05
† Information below the double lines is from observed patients younger than 12 years old
‡ Only explanations concerning occlusion therapy
§ For example: "You must...", "You need to...", "Remember that you…"
| For example: "the best is...", "it is important...", "I can imagine...", "try to…", "I advise you to…"
Other formulations used: "Do you have any questions?", "Is there something else that you want to know?"

occlusion therapy is very effective, an increase in visual acuity will only stagnate when compliance is very low. In an RCT (Stewart et al. 2007a) where children were prescribed either 6 or 12 occlusion hours per day, all children who patched more than 2 hours per day showed improvement in visual acuity, although children who patched more reached good visual acuity more rapidly. Children with moderate compliance will reach sufficient visual acuity in a longer period of time, and the orthoptist will only rarely suspect low compliance in such cases.

The allotted time for a patient's first visit was shorter in clinics located in non-native, low-SES areas than elsewhere (21′ vs. 27′24″). The allotted time for a follow-up visit (15′30″ vs. 16′44″), and mean time for explanation to the parent (2′30″ vs. 2′51″) and to the child (0′10″ vs. 0′24″), were also shorter in non-native, low-SES areas than elsewhere. Explanation to the child was short despite the fact that the orthoptists were being observed.

Children understand more about concepts of health and illness than is generally assumed (Holzheimer et al. 1998; Lewis et al. 1984; Tates et al. 2001). Several studies revealed that a more direct communication between physician and child contributes to an improved satisfaction with care and compliance to treatment, and to better health outcomes (Holzheimer et al. 1998; Loudon et al. 2006; Pantell et al. 1982; Tates et al. 2001).

Orthoptists never used the official medical interpreter, as they found it too time consuming. This has also been described in earlier studies about communication between non-native patients and general practitioners (Ramirez et al. 2008).

There were several limitations in this study. First, the presence of the researcher may have influenced the everyday behaviour of orthoptists during the 1-day observations, but it is unlikely that this had a major effect on our results because orthoptists were not aware of which exact items they were observed on and what behaviour we found desirable. Furthermore, we observed 15 consultations per orthoptist, on average, which is suggested to give a representative reflection of the orthoptists' activity (Pringle et al. 1990). Secondly, the questionnaires were not validated, which may have allowed for subjective interpretation of the results by the researchers. Finally, the second group of 23 orthoptists was not randomly selected, but volunteered for the study. This might explain their high interest in the subject and the acknowledgment that compliance is an important issue within the orthoptic practice.

Despite these limitations, we have been able to give an inventory of the conception, awareness, attitude, and actions to deal with noncompliance, which vary considerably among orthoptists. Orthoptists are aware that compliance is an important issue in their current practice, however, and are eager to learn more about structured methods to detect and to deal with noncompliance. It is, therefore, advisable that knowledge concerning compliance should be actively disseminated. Techniques to detect and to deal with noncompliance should be included into the orthoptic training curriculum and practice.

Acknowledgments

The Occlusion Dose Monitor was developed at the department for Medical Technical Development at the Academical Medical Centre Amsterdam, The Netherlands in 1996-1997 as a public domain project.

The authors thank the 32 participating orthoptists for their contribution to the study (names are listed per group, in alphabetical order of the clinic, and in alphabetical order of the names): Group A: Medical Centre Haaglanden, The Hague (E. van Minderhout, B. Simonsz - Tóth, M. Vermeulen-Jongen), Mesos Medical Centre, Utrecht (A. van de Bovenkamp - Scheper, A. Versteeg), Ruwaard van Putten Hospital, Spijkenisse (M. Kruiswijk), Sint Lucas Andreas Hospital, Amsterdam (A. Burger - Meywaard), Vlietland Hospital, Vlaardingen (S. Rousse, D. Valster). Group B: Amphia Hospital, Breda & Oosterhout (K. Croft, C. Klerks - Kuijpers, P. Neomagus, C. Stoop), Catharina Hospital, Eindhoven (E. van Rooijen - Troost), Flevo Hospital, Almere (L. Bakker, N. Bos - De Bruin), Isala Clinics, Zwolle (A. Wellner - Fokker), Kennemer Hospice, Haarlem (W. Jutte, A. Hupsch - Van den Heuvel, A. Spaan), Lievensberg Hospital, Bergen op Zoom (M. de Rooij, A. van Voorden - Van der Knaap), Máxima Medical Centre, Eindhoven & Veldhoven (E. Cockx - Huitema), Medical Clinic, Alkmaar (W. Hoogeveen, I. Huisman), Rivierenland Hospital, Tiel (T. Kooij - 't Spijker), Tergooi Hospitals, Hilversum (D. Hamers), Twenteborg Hospital, Almelo (I. 't

Hart-Leemhuis, M. Hazenkamp - Boekhold, F. Jansen, D. van der Nagel - Pronk), Swedish Red Cross Hospital, Zierikzee; Van Weel-Bethesda Hospital, Dirksland; & Medical Centre, Middelburg (J. Kieviet - De Geus).

The authors also wish to thank all 157 Dutch orthoptists for completing the questionnaire, G.J.J.M. Borsboom of the Department of Public Health, Erasmus MC University Medical Centre Rotterdam, for assistance with statistics, and Prof.dr. J. Passchier of the Department of Psychology and Education, Free University Amsterdam, for his advice.

Acknowledgement for proofreading and correcting the English edition goes to Mrs. K. Verhoef of VERT-ICAL Translation & Administration.

Chapter 5

Comfort of wear and material properties of eye patches for amblyopia treatment and the influence on compliance

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Strabismus. 2012; 20(1):3-10

ABSTRACT

Purpose. Occlusion therapy for amblyopia has been the mainstay of treatment for centuries, however, acceptance of the patch is often lacking. This study evaluated comfort of wear of the eye patch and assessed the mechanical properties in order to achieve a more individualised prescription.

Methods. For 8 consecutive days, parents used each of the 4 main brands of patches for 2 consecutive days in a randomised fashion. After 2 days a 21-item questionnaire was completed to evaluate comfort of wear for each patch. Compliance was measured electronically using the Occlusion Dose Monitor (ODM). In addition, breathing capacity at 23°C and 33°C, resistance to water penetration, opacity and strength of adhesion to the skin were measured.

Results. Twenty-four children participated. Overall, satisfaction was moderate: large differences in discomfort when removing the patch, skin reaction, and cosmetic appearance were found. In the material measurements large differences were found in opacity and strength of adhesion to the skin. In all brands breathing capability was minimal. Answers given by the parents matched the physical properties of the eye patch. There was no difference in electronically measured compliance between patches.

Conclusions. We found large differences in comfort of wear and mechanical properties. Therefore, when prescribing a certain brand of patch, the wide variety needs to be taken into account. Further study into these properties seems warranted; especially breathing capability requires improvement since children often wear them for a longer period of time. This could contribute to increasing satisfaction and consequently may improve compliance.

INTRODUCTION

Amblyopia is the most common visual defect in children with a prevalence of 3%-5% (Attebo et al. 1998). The condition can be partially or completely treated preferably before the age of six (Wiesel et al. 1963a). Traditionally, treatment involves glasses and / or occlusion of the nonamblyopic eye with an adhesive patch applied directly to the skin around the eye, thereby forcing the use of the amblyopic eye. Nowadays, a wide selection in brand of patches is available, with patches varying in size, colour, elasticity, and marketing policy.

Whether or not a patch adheres to the skin depends on a number of factors, e.g., skin type, the child's activities while wearing the patch, and adhesive strength of the patch. The eye patches are mostly made of nonwoven materials. Some manufactures assert their brand of patch is hypo-allergenic, indicating special glue was used to reduce the occurrence of allergies and itching. It has been reported that parents dislike the cosmetic appearance of their child wearing an eye patch. Some argue that wearing the eye patch is uncomfortable, causes irritation to the skin and leads to considerable distress for the child, outweighing benefits from improvement in vision (Holmes et al. 2003; Hrisos et al. 2004; Norman et al. 2003; Packwood et al. 1999; Parkes 2001; Snowdon et al. 1997b).

To date, no study has investigated comfort of wear of the various brands of eye patches together with electronic recording of compliance and with assessment of their material properties. This study investigated comfort of wear in correlation with compliance, using the Occlusion Dose Monitor (ODM) and a questionnaire. In addition, we assessed material properties of different brands.

METHODS

Study population

Our study population was a subset of the "Implementation of a Compliance Enhancing Programme" (ICEP) study, described in detail elsewhere (see Chapters 7 and 9). In summary, all children between 3 and 6 years of age, with newly diagnosed amblyopia (interocular difference in visual acuity of at least 0.2 logMAR, in case of refractive amblyopia an 18-week spectacle adaptation was taken into account) who received occlusion therapy for amblyopia as initial treatment were included in that study. It investigated whether an educational programme, which improves compliance with occlusion therapy, can be implemented in current orthoptic practice (see Chapters 7 and 9).

Study design

The study was designed as a single blind, randomised, trial. The four most prescribed brands of occlusive eye patches, at the time of the study, in The Netherlands were used: 3M (Opticlude boys & girls), Master-Aid (Ortopad boys & girls), Lohmann-Rauscher (Pro-Ophta) and BSN Medical (Coverlet S). For convenience for the child and its parents, it was chosen to test only one type of patch of each brand, as testing more types would be a reason for some parents not to participate.

Comfort of wear

Between November 2007 and January 2008, families participating in the ICEP study were contacted by telephone by the researcher to obtain verbal consent, and an appointment for a home visit was made. Prior to the home visit, each patient was randomised to a certain sequence in which the patches had to be worn. All participating children wore each of the four brands of eye patches for two consecutive days together with the ODM, which measured compliance electronically (Chopovska et al. 2005; Fielder et al. 1994). The researcher was not aware of the sequence in which the children wore the patches. The children had already been patching before this trial and therefore the children and parents were not blinded. The patch was supposed to be worn for as long as the orthoptist had prescribed per day. Figure 5.1 shows the design of this trial. After wearing one brand for two days, a 21-item, non-validated questionnaire, designed by a focus group and called the "Occlusion patch Comfort Questionnaire" (OCQ), was completed by the parents during a home visit or telephone call (Appendix VII). Items #4, #10, #11 and #19 were adapted from the "Amblyopia Treatment Index: Patching Questionnaire" developed and validated by Cole et al. (Cole et al. 2001). Items #1, #2, #7, #12, #13 and #14 were copied from a survey about patching which was used in the ICEP study (see Chapter 8). Twenty

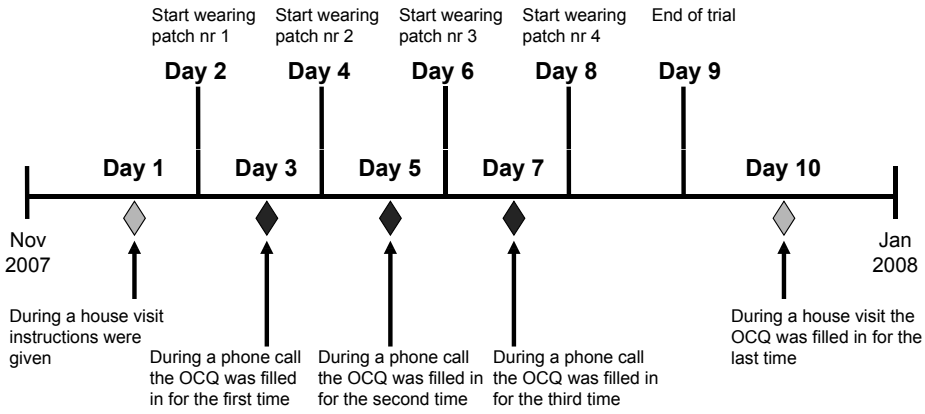

Figure 5.1) Timetable of the trial regarding wearing comfort and electronically measured compliance.

items were measured on a 5-point scale: a negative answer scored the most points. One question consisted of a visual analogue scale ranging from 0 till 10. Using this questionnaire we compared these patches regarding to: sizes, ease of removal, adhesive power, skin reaction, inconvenience during wearing, response of the environment, stress due to wearing the patch, overall opinion, and appearances.

The OCQ was filled out four times by the researcher (once for each brand); the first three times during a phone call and the fourth time during a home visit.

Measurements of material properties of the patch

The material properties of all the available types per brand at the time (2006) were tested (see also Table 5.2). Some types have been changed by the manufacturer since. The following material properties were investigated: breathing capacity, resistance to water penetration, opacity, and strength of adhesion to the skin.

Breathing capacity was tested at 23ºC (73ºF) and 33ºC (91ºF): the patches were glued on top of small plastic pots, each containing 20 grams of water. The pots were completely sealed off by the patch; the only way of ventilation was through the patch. They were left on a rocking table in a stove. The various patches were tested simultaneously at a temperature of 23ºC (73ºF) and a humidity of 30%. The same procedure was followed at a temperature of 33ºC (91ºF) and a humidity of 22%. These were standard temperatures used by TNO Enschede to test the breathing capacity of all types of textiles. To make sure

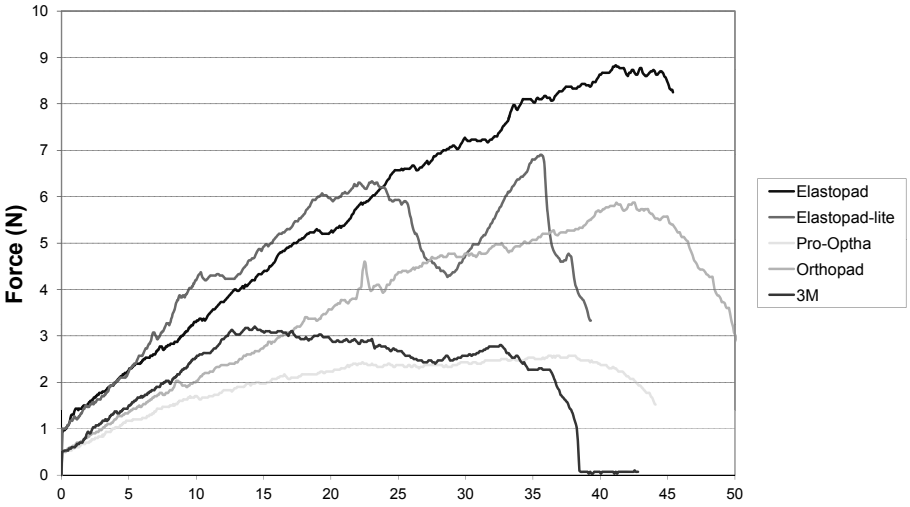

Figure 5.2) To remove the patches from the skin an average maximum force of 8.8 Newton (BSN Medical), 5.9 N (Master-Aid), 3.2 N (3M) and 2.6 N (Lohmann-Rauscher) was needed.

Table 5.1) A summary of the results regarding wearing comfort and compliance

Occlusive eye patches	Price per patch[1]	Compliance[2]	Opinion of the parent concerning all aspects (scale from 1-10)[3]	Available sizes	Size of the patch[4]	Adhesive power[5]	Painful removal?[6]	Skin reaction[7]	Inconvenience during wearing[8]	Reactions from others concerning the patch[9]	Appearance of the patch[10]	Stress due to wearing the patch[11]
	€	%	*			*	*	*			*	
3M (Opticlude boys & girls) N = 22	0.79-0.83	72.5	7.7	3	+	Strong	Some	Some	No	No	++	No
Master-Aid (Ortopad boys & girls) N = 22	0.86	88.2	6.9	3	+	About right	No	No	No	No	++	No
BSN Medical (Coverlet S) N = 23	0.80-1.11	77.5	6.0	3	+	Very strong	Some-severe	Severe	No	No	--	No
Lohmann-Rauscher (Pro-ophta) N = 21	0.74-0.93	86	5.9	2	+	Weak	No	No	No	No	--	No

++ = very good, + = good, ⊡ = fair, - = poor, -- = very poor

* = a significant difference between the four patches with $p < 0.05$

[1] Range of the price (based on contacting five pharmacies in Rotterdam)

[2] Electronically measured compliance in percentages: Opticlude Boys & Girls n = 14, Ortopad Boys & Girls n = 15, Coverlet S n = 15 and Pro-ophta n = 14

[3] "How would you rate this brand on a scale from 0 till 10, where 0 means very poor and 10 means excellent, when looking at all aspects of this patch" (#21)

[4] "The patch my child wore was?" (#1)

[5] "The patch sticks to the skin?" (#2), "How well did the patch stick on the eye of your child?" (#15)

[6] "How much pain did your child have when removing the patch?" (#3)

[7] "Wearing the patch makes my child's eyes or eyelids red or irritated" (# 4), "How red or painful was the skin of your child a few minutes after removing the patch?" (#5), "How red or painful was the skin of your child more than a hour after removing the patch?" (#6)

[8] "Was your child inconvenient during patching?" (#8), "My child does not seem to mind wearing the patch once it is on" (#19)

[9] "How often is your child getting negative reactions from others when he/she is wearing the patch?" (#9), "I notice that other children stare at my child when the patch is on" (#10)

[10] "My child thinks the patch looks pretty" (#20)

[11] "My child gets upset because of patching" (#12), "I get upset because of patching" (#13), "Other family members get upset because of patching" (#14)

these conditions were constant, a separate digital thermometer was used to measure the temperature and humidity. After 24 hours the amount of water left in the plastic pot was measured. The breathing capacity, or 'water vapour resistance' (Ret), was calculated using: $m \times Pa/W$ ($m = \pi \times r^2 \times$ time [seconds]; Pa = saturated [water] vapour pressure, depending on temperature and humidity; W = difference in amount of water (grams) after a certain time, with 1 gram of water equalling 2430 Joule). The Ret is classified as follows (references obtained from The Netherlands Organisation for Applied Scientific Research, Textile Industry): Ret > 40: 'uncomfortable', e.g. raincoat and has a restricted wearing time; $20 <$ Ret ≤ 40: 'somewhat comfortable'; Ret ≤ 20, which equals 'comfortable'.

Resistance to water penetration (for the purpose of simulating rainy weather conditions) was tested as follows: one drop of water was placed in the centre of the front side of the patch. To ensure an equal amount of water on each patch, one drop of water was chosen. The time necessary for the drop to be absorbed was measured with a stopwatch. Material was considered water resistant when the drop of water was not absorbed within 1 minute.

Opacity of the patches was tested by gluing all patches to a fluorescent lamp of 18 watts. We measured the amount of light transmitted through the centre and at the side of the patch. 100% light transmission equals 'no patch present'.

Strength of adhesion of the patch to the skin was tested using the 'maximum force grab method'. The patches were stuck to the skin and the force necessary to pull the patch was measured (expressed in Newton).

The breathing capacity was tested at the laboratory of the ErasmusMC University Medical Centre Rotterdam; the other tests were performed at The Netherlands Organisation for Applied Scientific Research (TNO), Textile Industry, Enschede.

Statistical analysis

Comfort of wear was analysed using ordinal regression analyses (with the answer to the question as outcome variable and the patch and child as categorical explanatory variables), with odds ratios as effect sizes. This was done for items #1 to #20. Item #21 ('How would you rate this brand on a scale from 0 till 10, where zero means very poor and ten means excellent, when looking at all aspects of this patch.') consisted of a visual analogue scale ranging from zero till ten: parents gave an overall grade for each of the four patches. Means and *P*-values of the four brands were calculated using a univariate general linear model with question number 21 as outcome variable and the brand and child as factors.

Compliance was measured electronically during eight days. It was defined as the actual occlusion time measured with the ODM divided by the prescribed occlusion time and expressed as a percentage. Differences in compliance between the four groups of patches were assessed using a univariate general linear model with compliance in percentages as dependant variable and the brand and child as categorical explanatory variables. $P < 0.05$ indicated statistical significance.

RESULTS

Study population

Twenty-four children were contacted of whom 22 fully completed the trial. One child withdrew after one day because removal of the first patch was too painful. Another child became ill and could therefore not finish the trial. Before the study commenced, 77% used 3M, 11% Master-Aid, 1% BSN Medical and nobody used Lohmann-Rauscher. This had been prescribed by the treating orthoptist. Mean prescribed occlusion time was 145.0 minutes per day (SD 74.8; range 60-300). Mean age was 4.8 years (SD 0.989) and 58.3% were boys. Overall mean compliance, as measured with the ODM, was 80.95% (range 72.5-88.2).

Comfort of wear

The results regarding comfort of wear are summarised in Table 5.1. Item #7 ('Has patching got a negative effect on the relationship between you and your child?') and #11 ('I have trouble keeping the patch on my child') were omitted from analyses because they were answered uniformly (no differences were found between the four brands). Items #16 ('How well did the patch stick on the eye of your child during sportive activities, such as cycling, running, playing etc?'), #17 ('How well did the patch stick on the eye of your child when it was warm or when he / she was sweating?') and #18 ('When your child also wears glasses: how well can the glasses be combined with the patch, without the ODM being attached?') were also omitted because the answer 'not applicable' was given more than 50%.

Prices did not differ between the patches and varied between □0.74-0.93 per patch (Table 5.1, second column). Compliance was not significantly influenced by the brand of patch ($P = 0.179$). See Table 5.1, third column.

There was a significant difference in the overall score for each patch ($P \leq 0.05$), which concerned all aspects of the patches; this score ranged from 0 (very bad) to 10 (very good) with 3M (Opticlude boys & girls) receiving the highest score. 3M (Opticlude boys & girls),

Master-Aid (Orthopad boys & girls) and BSN Medical (Coverlet S) all had 3 sizes available; Lohmann-Rauscher (Pro-ophta) had 2 sizes available. The opinion of the parents about the sizes did not differ between the brands (Table 5.1, fifth and sixth column).

Statistical significant differences were also found in the perception of the parents regarding adhesive power (Table 5.1, seventh column shows the results of questions 2 and 15, because these results were similar). 3M (Opticlude boys & girls) gave some and BSN Medical (Coverlet S) a severe skin reaction. Master-Aid (Orthopad boys & girls) and Lohmann-Rauscher (Pro-ophta) did not give a skin reaction (Table 5.1, 9th column shows #4, #5 and #6 combined, because these results were similar). The children were not disturbed more by any particular kind of patch (Table 5.1, 10th column). Reactions from others (family, friends etc.) were not significantly different among the four brands (Table 5.1, 11th column). We did find a statistical significant difference in the opinion of the parents and children regarding appearances: 3M (Opticlude Boys & Girls) and Master-Aid (Orthopad boys & girls) scored very high, BSN Medical (Coverlet S) and Lohmann-Rauscher (Pro-Ophta) scored very low (Table 5.1, 12th column). The level of stress experienced by their children, as reported by the parents, did not differ between brands (Table 5.1, 13th column). The majority of the parents (86.7%) preferred incisions at the side of the patch to no incisions; only 3M (Opticlude boys & girls) and BSN Medical (Coverlet S) had incisions at the side of the nose, the other two brands had not. However, all these children wore glasses.

Three of the 22 children who completed the trial switched to another brand after the trial.

Material properties

The results of the water vapour resistance test showed that none of the various brands of patches were 'comfortable' to wear at a temperature of 23˚C (73ºF) (equivalent to Ret < 20; Table 5.2). BSN Medical (Coverlet S) and 3M (Opticlude) were actually 'very uncomfortable' to wear (Ret > 40). At a temperature of 33ºC (91ºF) and a humidity of 22%, Lohmann-Rauscher (Pro-Ophta) and Master-Aid (Orthopad) were 'comfortable' to wear and BSN Medical (Coverlet S) and 3M (Opticlude) were 'somewhat comfortable'. All brands, except Master-Aid (Orthopad), were water-resistant.

Only one brand was able to eliminate more that 70% of the light transmitted by a fluorescent lamp (BSN Medical), whereas other brands eliminated 50% (Master-Aid), 48% (Lohmann-Rauscher), and 20% (3M) of the light.

To remove the patches from the skin an average maximum force of 8.8 Newton (BSN Medical), 5.9 N (Master-Aid), 3.2 N (3M) and 2.6 N (Lohmann-Rauscher) was needed (Figure 5.2).

Table 5.2) Physical properties, flexibility, hygiene, sizes, and distributed gadgets or gifts of the 4 eye patches.

Brand / Type	*1) Breathing capability (Ret)		*2) Opacity centre of patch	*3) Water resistant	Force to remove patch from skin (Force maximum; N)	Flexibility / Elasticity
	23° C	33° C				
3M Opticlude					3,2	breadthwise only, very limited
Skin coloured	64.1	34.9	89%	> 1 min		
Blue			79%	> 1 min		
Red			82%	> 1 min		
Green			78%	> 1 min		
Master-Aid Ortopad					5.9	breadthwise only
Regular - skin	21.8	15.2	49%	10-15 sec		
Regular - white			54%	> 1 min		
Simpathy - red			42%	< 1 sec		
Sympathy - black/ white			51%	black 10 sec; white > 1 min		
Simpathy - blue			43%			
Lohmann-Rauscher						
Pro-ophta	30.1	14.5	62%	> 1 min	2.6	lengthwise only
BSN Medical						
Elastopad	53.9	29.7	19%	> 1 min		breadthwise only
Elastopad - Lite	39.9	21.8	29%	> 1 min		breadthwise only
Coverlet - S	55.7	33.3		> 1 min		
Coverlet - S with sticker	70.8	37.8				very flexible in all directions

Mechanical properties, flexibility, hygiene, sizes, and distributed gadgets or gifts of the 4 eye patches. *1) Ret: water vapour resistance and classified as follows: Ret > 40 = uncomfortable (e.g. raincoat) and has a restricted wearing time 20 < Ret < 40 somewhat comfortable
Ret < 20 = comfortable to wear
*2) Opacity: 100% equals 'no patch present'
*3) Material is labelled water-resistant when the drop of water is not absorbed within 1 minute

DISCUSSION

This is the first randomised trial investigating the mechanical patch properties in relation to comfort of wear and electronically measured compliance. The tests performed demonstrated large differences in the patch properties between the four brands. Breathing property of all patches was minimal at room temperatures; some patches could be compared to wearing a raincoat in the sun and would therefore clearly be more suited when occluding for shorter periods of time. There was also considerable difference in strength necessary to remove the patch from the skin. The maximum force required to remove a patch from the skin varied between 2.6 (Lohmann-Rauscher) and 8.8 (BSN Medical) Newton, clearly demonstrating that certain brands of patches would be more suitable for longer duration of patching. No patch was able to eliminate 100% of the light, however, it is unclear whether the patch must exclude all light and form, or if it is sufficient to exclude form, but allow the passage of (some) light.

We could not correlate electronically measured compliance to a certain brand. This might be due to the small sample size of the study. Also, for future study, skin type and activities during patch wear could be taken into account.

Answers given by the parents were comparable to the measured mechanical properties of the patch. Overall, parents were moderately satisfied with the eye patches. As most of the families used 3M or Master-Aid prior to the study, this may have influenced the answers to the questionnaire.

Considering these results we suggest the force necessary to remove the patch caused by the glue layer, which causes irritation of the skin, to be the most important factor influencing comfort of wear. Therefore, it seems reasonable to expect orthoptists to take comfort of wear into consideration when prescribing a certain brand of patch and for manufacturers to spend more time and effort on improving the properties, especially the glue layer of their patches.

Acknowledgments

The authors thank Elizabet Vukovic, Elizabeth van de Graaf, Bregje van Dijk, Ellen van Minderhout, Marleen Vermeulen, Roberta Delle Site, Janine van Zon, Eline Deelstra and Jorrien Bouma for their help with data collection and efforts in recruiting the children. We also want to thank all orthoptists for their effort in participating in this study. We thank The Netherlands Organization for Health Research and Development for their grants. Finally, we are very grateful for the hospitality and help we received from A.W. Wypkema, Netherlands Organisation for Applied Scientific Research (TNO), Textile Industry, Enschede.

The Occlusion Dose Monitor was developed at the department for Medical Technical Development at the Academical Medical Centre, Amsterdam, The Netherlands in 1996-1997 as a public domain project.

Chapter 6

Sociocultural and psychological determinants in migrants for noncompliance with occlusion therapy for amblyopia

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Graefes Arch Clin Exp Ophthalmol. 2011;249(12):1893-1899

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ABSTRACT

Purpose. Compliance with occlusion therapy for amblyopia in children is low when their parents have low level of education, speak Dutch poorly, or originate from another country. We determined how sociocultural and psychological determinants affect compliance.

Methods. Included were amblyopic children between the ages of three and six, living in low socio-economic status (SES) areas. Compliance with occlusion therapy was measured electronically. Their parents completed an oral questionnaire, based on the "Social Position & Use of Social Services by Migrants and Natives" questionnaire that included demographics and questions on issues like education, employment, religion and social contacts. Parental fluency in Dutch was rated on a five-point scale. Regression analysis was used to describe the relationship between the level of compliance and sociocultural and psychological determinants.

Results. Data from 45 children and their parents were analyzed. Mean electronically measured compliance was 56 SD 44 percent. Children whose parents had close contact with their neighbours or who were highly dependent on their family demonstrated low levels of compliance. Children of parents who were members of a club and who had positive conceptualizations of Dutch society showed high levels of compliance. Poor compliance was also associated with low income, depression, and when patching interfered with the child's outdoor activity. Religion was not associated with compliance.

Conclusions. Poor compliance with occlusion therapy seems correlated with indicators of social cohesion. High social cohesion at micro level, i.e. family, neighbours and friends, and low social cohesion on macro level, i.e. Dutch society, are associated with noncompliance. However, such parents tend to speak Dutch poorly, so it is difficult to determine its actual cause.

INTRODUCTION

Amblyopia, a "lazy eye", is a preventable, unilateral loss of vision and affects approximately 3–4% (Attebo et al. 1998) of the population. Recovery is possible by occluding the better eye with a patch for several hours a day, generally before the age of six (Holmes et al. 2006a). Despite amblyopia treatment, approximately one-third of the affected children do not attain a visual acuity in the amblyopic eye to be able to read (0.5 decimals (Rahi et al. 2002a)). This is primarily caused by poor compliance with occlusion therapy, i.e., the eye is not patched according to the orthoptist's prescription (Loudon et al. 2006; Smith et al. 1995). In a previous study, we found that noncompliance with occlusion therapy correlated strongly with the parents' country of origin, poor parental fluency in the national language, and low levels of education (Loudon et al. 2006). It may be possible, however, that other factors related to the country of origin, such as religion, cultural integration, perspective on health, and culture, also play a role in the degree of compliance.

Immigrants encounter several difficulties once they arrive in the host country. They may not speak the language of the host country, may have had limited education, may be in a poor financial state, may be unemployed, may lack suitable accommodation, often have no social network, may have different cultural norms and religious beliefs; they must comply with new laws, as well as cope with possible racism and other exclusionary behaviour. These difficulties can be problematic when visiting a physician, resulting in miscommunication and possible noncompliance with treatment (Harmsen et al. 2003; Loudon et al. 2006; Norredam et al. 2009; Schouten et al. 2006; Van Wieringen et al. 2002). In addition, immigrant patients' views on health and disease may differ from those of native patients (Norredam et al. 2009; Schouten et al. 2006; Seeleman et al. 2005; Van Wieringen et al. 2002).

In The Netherlands, approximately one-fifth of the total population is non-native (Statistics Netherlands 2008). In Amsterdam, 49% of the population is of non-native origin; in Rotterdam and The Hague, 47% is non-native, and 31% of the population in Utrecht is non-native (Statistics Netherlands 2008). Most non-native inhabitants of these cities are first-generation immigrants, and live in ethnically diverse, low-SES, suburban neighbourhoods (Statistics Netherlands 2008). Among these inhabitants, immigrant-related determinants other than country of origin, fluency in Dutch and level of education may have an effect on the level of compliance with occlusion therapy. For these reasons, we explored whether sociocultural, psychological and differences in acculturation similarly affect compliance with occlusion therapy. We performed this study in ethnically diverse, low-SES, suburban areas of the cities Utrecht and The Hague.

METHODS

The current study was part of a nation-wide pre-post implementation study of compliance-improving measures (see Chapters 7 and 9), including an educational booklet, containing a wordless cartoon story on the importance of wearing the eye patch and a calendar with reward stickers for the children, and an information leaflet in six languages (Dutch, English, French, German, Turkish and Arabic) for the parents (Loudon et al. 2006). Nine orthoptists working at an orthoptic department in an ethnically diverse, low-SES, suburban area in one of the four major cities in The Netherlands (Amsterdam, Utrecht, The Hague and Rotterdam) were asked to recruit newly diagnosed amblyopic children between the ages of 3 and 6 years with a visual acuity difference of ≥1 logMAR line between the eyes and an amblyopic factor, and who were to undergo occlusion therapy for the first time.

Compliance was electronically measured by using the Occlusion Dose Monitor (ODM) (Chopovska et al. 2005; Fielder et al. 1995; Moseley et al. 1995). The ODM is a device that records, every 2 minutes, the difference in temperature between the front and back of the ODM. The parents were asked to attach the ODM to the front of the eye patch with double-sided adhesive tape; hence, the temperature at the back of the device was higher than at its front when the patch with ODM was worn on the eye (Loudon et al. 2006).

For this study, parents of children who received standard treatment (without the compliance-improving measures), living in Utrecht and The Hague, were asked to participate in an oral interview at their home: these interviews were conducted by two members of the research team (HA, FZ).

During the home visit, an extended questionnaire was applied. This structured oral questionnaire is based on a questionnaire which had been developed by The Netherlands Institute for Social Research /SCP and the Institute for Sociologic–Economic Research (ISEO) of the Erasmus University in Rotterdam. This questionnaire had been developed for the research project called SPVA (a Dutch acronym for "Sociale Positie en Voorzieningengebruik Allochtonen en Autochtonen" meaning "Social Position & Use of Social Services by Migrants and Natives") (The Netherlands Institute for Social Research 2007). Its purpose was to investigate the proportion of inhabitants of different ethnic minorities participating in the Dutch society, educational system and labour market, and to monitor these individuals over a period of several years (Groeneveld et al. 2003; Weijters et al. 2003). Approximately 4,200 households had filled out this questionnaire each year in the years 1988, 1992, 1994, 1996, 1998, 2002 and 2003.

For the current study, we used 172 of the 265 questions of the 2002 version of the SPVA questionnaire: questions concerning the educational level and labour of grandparents, aunts and uncles of the child, and questions about previous jobs, periods of unemployment or previous social payments of the parents were omitted. The remaining 172 questions concerned ten domains (Appendix VIII): *Demography, Migration & Household* (e.g., "What is your marital status?"); *Education* ("What is your highest level of education?"); *Employment* ("What is your current work?"); *Income* ("What is your gross family income?"); *Health* ("How is your health in general?"); *Language Usage* ("Do you experience difficulties with reading, for example, Dutch magazines?"); *Religion* ("How frequently do you visit religious communities?"); *Family Bonds* (Statement: "You can always count on your family."); *Social Contacts* ("Do you have Dutch neighbours / friends who visit you once in a while?"); and *Cultural Integration & Conceptualization* (Statement: "The Dutch society is open toward foreign people"). An additional domain *Lazy Eye* was added to the questionnaire, comprising 33 questions from "The Utility Analysis of Amblyopia" Questionnaire (Van de Graaf et al. 2004), "Children's Visual Function Questionnaire (VFQ) (Felius et al. 2004) and "Patching Questionnaire" (Cole et al. 2001) (Appendix VIII). Most questions could be answered on a 5-point scale ranging from 'Strongly Agree' to 'Strongly Disagree'. The questionnaire was presented to the parents in a face-to-face interview format.

During the home visit, the researchers rated the parent's fluency in Dutch on a five-point-scale (Loudon et al. 2006), with 1 'no fluency'; 2 'scarcely fluent / poor fluency'; 3 'moderate fluency'; 4 'good fluency'; and 5 'near native speaker/exceptional fluency'. The living conditions of the child were observed using a structured observation list to score the quality of the residence. This list was based on a score sheet used by the Ministry of Housing, Spatial Planning and the Environment to define the quality of a residence (Veenman et al. 1994). It contained questions such as: on what floor is the residence located, is there an elevator in the building, how many rooms are there in the apartment/house, and how many persons live at the residence. A home visit lasted approximately 2.5 hours.

Statistical Analysis

The initial visual acuity was the visual acuity as measured during the child's first visit to the orthoptic department. Visual acuity was measured using the Landolt-C chart; in younger children (3–4 years) a non-standardized picture chart or the E-chart was used. As non-standardized picture charts were used, visual acuity is indicated in logMAR instead of absolute values.

The questions, the electronically measured compliance scores obtained from the implementation study, and parental fluency in Dutch were analyzed in SPSS 16.0 for Windows. With univariate general linear regression model analysis, correlations between

independent variables, that is, all questions from the questionnaire, and the electronically measured compliance were determinant. Variables with statistical significance were further analyzed in multiple regression analysis. A *P*-value of <0.05 indicates statistical significance.

RESULTS

A total of 114 children were recruited in the pre-implementation phase of the study in Amsterdam, Rotterdam, Utrecht and The Hague. Of these, 33 lived in Amsterdam or Rotterdam, and, were not eligible because of long travel distances. Parents of 14 other children could not be reached despite two phone calls and two house calls. Fifteen parents refused to participate in the oral interview: the reason they all gave was that they had no time. Of the 52 parents who responded to the interview, 24 had a 'moderate' or 'poor' level of fluency in the Dutch language. Sixteen parents were of Dutch origin, 13 Moroccan, 13 Turkish, and ten had other countries of origin. Seven were Christians, 30 were Muslims, seven had no religion and eight had other religious affiliations. The mean age of the children was 4.84 SD (standard deviation) 1.24 years when occlusion therapy was started. The mean prescribed occlusion time was 3:37 SD 1:15 hours a day, for 7 days a week, at the initiation of occlusion therapy. The mean initial visual acuity was 0.42 SD 0.33 logMAR in the amblyopic eye and 0.07 SD 0.11 logMAR in the better eye.

Data on compliance was available for 45 of the 52 children: in seven cases, data were incomplete due to repeated failure of the ODM. Mean compliance was 56 SD 44 percent. Compliance followed a bimodal distribution (solid polynomial line in Fig. 6.1) with two peaks: one at 90%, the other at 0%. Twenty-two of the 45 children occluded less than half of the occlusion time prescribed by the orthoptist (Fig. 6.1).

Fifty-five of the 205 questions were excluded from analysis: 34 did not apply to or were not answered by the majority of parents and 21 were answered unanimously, i.e., all parents answered 'agree' or 'disagree'. Univariate analysis showed that 18 of the remaining 150 variables correlated significantly with the electronically measured compliance. These variables belonged to the domains *Income, Health, Family Bonds, Social Contacts, Cultural Integration & Conceptualization,* and *Lazy Eye*. Table 6.1 depicts the 18 variables that correlated significantly with the electronically measured compliance.

In the domain *Income*, low income (i.e., < □1,750 net/month, the average income in The Netherlands (Economic Policy Analysis Netherlands 2009)) was associated with poor compliance (P = 0.017).

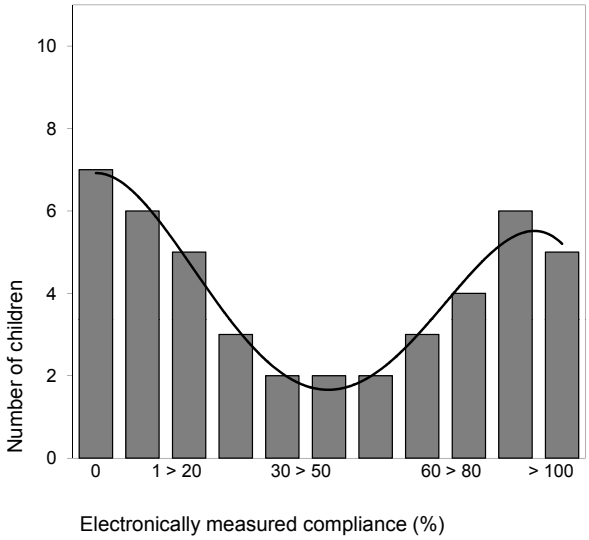

Figure 6.1) Bar chart of electronically measured compliance in the study population (n = 45). Compliance was defined as the actual occlusion time measured with the ODM divided by the prescribed occlusion time and expressed as a percentage, in which 100% indicates a compliance of 100%. This graph followed a bimodal distribution with two peaks (solid line).

In the domain *Health*, children of parents who had experienced periods of depression during the previous four weeks had a significantly lower compliance (P = 0.022).

In the domain *Family Bonds*, two questions were associated with compliance: parents who agreed that grandparents should be prepared to babysit their grandchildren on a regular basis, and who agreed that grown-up children, who lived near their parents, should visit them at least once a week, had a lower compliance (P = 0.001 and P = 0.039 respectively).

Four questions in the domain *Social Contacts* demonstrated correlations with compliance: parents who had frequent close contact with neighbours demonstrated poor compliance. Children of parents who were members of a social club, for example, a sports club or a religious organization had high compliance (P = 0.003). Parents of 19 children were member of one club; five parents were member of two or more clubs. Twelve parents were member of a religious organization, ten of a sports club, three of a music association or a theatrical company, two of a school association, one of a labour union, and one of a political organization.

In the domain *Cultural Integration & Conceptualization* in Table 6.1, eight questions pertaining negative perceptions of the Dutch society correlated with low compliance: parents who agreed with these questions were significantly less compliant (30%-40%)

Table 6.1) In univariate analysis, 18 questions were associated with electronically measured compliance with occlusion therapy. On the left, in italics, all domains are listed. All questions that were not significant were omitted. Mean electronically measured compliance in percentage (in brackets is N) per response on each question is given. * P < 0.01

Domain	Mean Compliance in % (N)					P-value
Demography, Migration & Household						
Education						
Employment						
Income	*Yes*	*No*				
Family income of <€1,750.00 net/month	44 (27)	77 (14)				0.017
Health	*Yes*	*No*	*Not answered*			
In the past 4 weeks, have you experienced any periods of depression?	36 (11)	59,1 (30)	82 (4)			0.047
Language Usage						
Religion						
Family Bonds	*Strongly agree*	*Agree*	*Agree/ disagree*	*Disagree*	*Strongly disagree*	
"Grandparents should be prepared to babysit their grandchildren on a regular basis"	27 (9)	45 (14)	64 (10)	82 (12)	- (0)	0.001*
"Grown-up children, who live near their parents, should visit them at least once a week"	38 (9)	54 (26)	70 (6)	86 (4)	- (0)	0,039
Social Contacts	*Very well*	*Good*	*Moderate*	*Poor*	*None*	
How well do your neighbours interact with one another?	31 (11)	63 (33)	94 (1)	- (0)	- (0)	0,022
	Very unfortunate	*Unfortunate*	*Do not matter*	*Positive*	*Very positive*	
How would you feel if your neighbours moved away?	30 (7)	52 (24)	74 (14)	- (0)	- (0)	0,024
	As little as possible	*Greet in passing*	*Talk once in while*	*Visit once in a while*	*Frequent contact*	
To what degree should neighbours have contact with one another?	86 (2)	74 (1)	63 (20)	51 (12)	39 (10)	0,019
	Yes	*No*	*Not answered*			
Are you a member of a club?	72 (25)	37 (19)	2 (1)			0.003*

Cultural Integration & Conceptualization	*Strongly agree*	*Agree*	*Agree/ disagree*	*Disagree*	*Strongly disagree*	
"It is more important for boys to earn their own money than for girls"	- (0)	32 (11)	44 (3)	56 (8)	68 (23)	0,020
"Elderly people should be able to live with their children"	32 (8)	51 (22)	70 (11)	88 (4)	- (0)	0,014
"Parents should allow their grown-up children to live in their home"	31 (6)	49 (19)	67 (17)	85 (3)	- (0)	0,026
"Nowadays, people are getting divorced too easily"	30 (6)	44 (12)	58 (12)	72 (15)	- (0)	0,023
"Elderly family members have more to say when making an important decision than do younger ones"	- (0)	41 (17)	54 (6)	66 (20)	79 (2)	0.005*
"In the Netherlands, people speak too openly about sexuality"	34 (6)	46 (15)	58 (8)	71 (15)	83 (1)	0,033
"Certain sectors of the economy only keep going because of non-native employees"	32 (6)	49 (20)	65 (13)	81 (6)	- (0)	0,026
	Yes	*A little*	*No*	*Absolutely not*		
"Dutch people are reticent"	29 (5)	48 (20)	67 (17)	86 (3)		0,021
Lazy Eye	*Strongly agree*	*Agree*	*Agree/ disagree*	*Disagree*	*Strongly disagree*	
"My child has difficulty playing outside when he/she wears the patch"	0 (1)	- (0)	29 (6)	50 (16)	22 (70)	0.007*
	(Almost) never	*Occasionally*	*Sometimes*	*Often*	*(Almost) always*	
"My child is less active when he/she is being treated"	66 (24)	51 (11)	36 (9)	21 (1)	- (0)	0.046

than parents who did not agree with these questions. For example, parents who were of the opinion that it is more important for boys to earn their own money than for girls, that people are getting divorced too easily nowadays and that, in The Netherlands, people speak too openly about sexuality, had a significantly lower compliance (P = 0.020, P = 0.023, and P = 0.026 respectively). Poor compliance was also found in parents who found that elderly people should be able to live with their children, that parents should allow their grown-up children to live in their home, and that the elderly family members have more to say when making an important decision than younger family members (P = 0.014, P = 0.026 and P = 0.005, respectively).

In the domain *Lazy Eye*, low compliance correlated with the parental opinion that the child had difficulty with playing outside while wearing the patch (P = 0.007). When parents said the child was less active while wearing the patch, compliance was significantly lower (P = 0.046).

No questions in the domain *Religion* were found to be associated with compliance. The kind of religion, differences in interest in religion, frequency of practicing the religion, frequency of visiting religious communities (e.g., the church or mosque), and strict or less strict religious beliefs did not affect compliance with occlusion therapy.

The 18 variables that were significant in univariate analysis were included in the multiple regression analysis. One variable remained significant: "*In The Netherlands, people speak too openly about sexuality*" (P = 0.002).

DISCUSSION

In this group of children from ethnically diverse, low-SES areas, we found that compliance correlated with the degree of integration within Dutch society. Low levels of compliance were associated with close family bonds and close neighbour contacts. Conversely, parents who were member of a club and who had positive conceptualizations of Dutch society had good compliance. Low compliance was also found in case of low income or depression of the parents and when patching interfered with the outdoor activity of the child. Almost half of the children with amblyopia from these ethnically diverse, low-SES areas occluded less for than half of the prescribed occlusion time.

Perception of health status and the utilization of health services differ between native and non-native inhabitants (Fassaert et al. 2009; Kunst et al. 2007; Norredam et al. 2009; O'Malley et al. 1999; Uiters et al. 2006). O'Malley et al. (1999) and Fassaert et al. (Fassaert et al. 2011) found that participation of migrants in their host culture is associated positively with more the use of health care services. Fassaert et al. (Fassaert et al. 2009) found that Turkish migrants in The Netherlands who spoke Dutch more fluently consulted medical specialists less often, but utilized mental health services more often. Higher levels of social interaction with Dutch people was related with more utilization of mental health services among Turkish women (Fassaert et al. 2009). However, other studies reported a high utilization of general practice services among patients with low-SES and by non-native patients (Kunst et al. 2007; Norredam et al. 2009; Uiters et al. 2006).

In our study, we found a relationship between compliance and the degree of social cohesion within a community or society. Social cohesion describes the degree to which human behaviour expresses commitment and solidarity within a given community (Schnabel et al. 2008). The degree of social cohesion is linked to social confidence and social trust, is inversely associated with social exclusion, and defines a person's social position within a society (Schnabel et al. 2008). Our findings suggest that the children of parents who have close family bonds and close neighbour contacts (more socially cohered at the micro level) had a significantly lower mean electronically measured compliance. Parents who were members of a club and who had positive conceptualizations of Dutch society (more socially cohered at a macro level) had, in general, good electronically measured compliance.

It is difficult to determine the exact cause of noncompliance (Schouten et al. 2006), as the inability to speak Dutch necessitates family bonds and neighbour contacts. Secondly, parents who do not speak Dutch misunderstand the orthoptist's instruction (Harmsen et al. 2003; Loudon et al. 2006; Van Wieringen et al. 2002). Similarly, poor fluency in Dutch decreases the chance to find work. Poverty may induce depression (Gonzalez-Castro et al. 2011). Depressive moods affects social cohesion (Fassaert et al. 2011).

We found no evidence that the kind of religion, frequency of practicing the religion, or strict or less strict religious beliefs, had any effect on compliance with occlusion therapy. The role of religion on health care behaviour is still unclear: religious behaviour may influence health care behaviour positively or negatively. In a study among HIV patients, certain religious practices were positively associated with treatment adherence, whereas other religious beliefs played a negative role due to the stigma attached to HIV disease (Parsons et al. 2006). An earlier report about patients with diabetes mellitus in Sweden (Hjelm et al. 2003) suggested that religious differences indirectly affect the degree of self-care behaviour. In that study, Swedes had active self-care behaviour and a healthy and controlled life-style, whereas immigrants from (former) Yugoslavians and Muslims emphasized enjoyment of life and a passive self-care attitude. Although Muslims tended to take their diabetes as 'the will of Allah or God', they searched more actively for information about management of diabetes (Hjelm et al. 2003).

Our population of parents who lived in ethnically diverse, low-SES areas is not representative to the general population in The Netherlands. Our study was restricted to 45 children. However, our data collection was highly comprehensive, involving the distribution and recollection of the ODMs and the administration of the comprehensive oral questionnaire, including 205 questions, during home visits. On the other hand, the oral administration of the questionnaire resulted in a 100% response. In case of a postal questionnaire, the

response would have been much lower, not only because of the lengthy questionnaire, but also because some of the participants were not able to read. In case of poor fluency in Dutch, parents were asked to arrange a relative of friend to interpret the questions. In some instances, an informal interpreter had to be approached on the spot (Turkish parents excluded, because both researchers spoke Turkish fluently). Moreover, the researchers tried to minimize language errors by paraphrasing and reconfirming the parents' answers, where necessary.

In conclusion, poor compliance with occlusion therapy was correlated with the degree of integration within the Dutch society and indicators of social cohesion. High social cohesion at micro level, i.e., family, neighbours and friends, and low social cohesion on macro level, i.e., Dutch society, were associated with noncompliance. We do not expect, however, that these findings will cause a change in orthoptic clinics, but it may support orthoptists working in ethnically diverse, low-SES areas in better understanding the compliance behaviour of non-native patients. Finally, we believe that good cultural integration of immigrants within the society of their host country seems to be of great importance in achieving better compliance with medical treatment, in general.

Acknowledgments

A part of this study was the graduate thesis of Ms. H. Akcan and Ms. F. Ziylan of the HU University of Applied Sciences Utrecht (Hogeschool Utrecht) The Netherlands, Department of Orthoptics in October 2008.

The Occlusion Dose Monitor was developed at the department for Medical Technical Development at the Academical Medical Centre, Amsterdam, The Netherlands in 1996–1997 as a public domain project.

The authors thank Cees Zandvliet for the explanation about the SPVA study, Ellen van Minderhout, Marleen Vermeulen-Jongen, Brigitte Simonsz-Tóth, Arieneke Burger-Meywaard, Marlien Kruyswijk, Sylvia Rousse and DorienValster for their help with data collection and efforts in recruiting the children.

Acknowledgement for proofreading and correcting the English edition goes to Mrs. K. Verhoef of VERT-ICAL Translations & Administration and Mrs. A. Van Rij–De Jong.

Chapter 7

An educational cartoon accelerates amblyopia therapy and improves compliance, especially among children of immigrants

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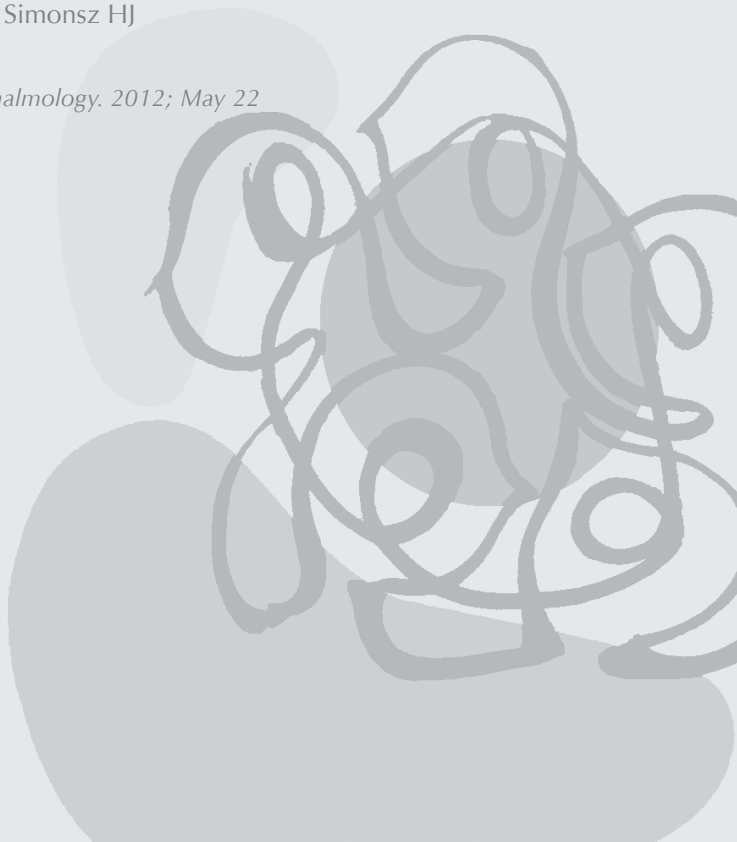
Accepted for publication by Ophthalmology. 2012; May 22



ABSTRACT

Purpose. We showed previously that an educational cartoon that explains without words why amblyopic children should wear their eye patch improves compliance, especially in children of immigrant parents who speak Dutch poorly. We now implemented this cartoon in clinics in low socioeconomic status (SES) areas with a large proportion of immigrants and clinics elsewhere in The Netherlands.

Design. Clinical, prospective, non-randomised, pre-implementation and post-implementation study. **Participants.** Amblyopic children aged 3 to 6 years who started occlusion therapy. **Methods.** Pre-implementation, children received standard orthoptic care. Post-implementation, children starting occlusion therapy received the cartoon in addition. At implementation, treating orthoptists followed a course on compliance. In low-SES areas, compliance was measured electronically during one week. **Main outcome measures.** The clinical effects of the cartoon - electronically measured compliance, outpatient attendance rate, and speed of reduction in intraocular-acuity difference (SRIAD) - averaged over 15 months of observation.

Results. In low-SES areas, 114 children were included pre-implementation versus 65 children post-implementation; elsewhere in The Netherlands, 335 versus 249 children were included. In low-SES areas, mean electronically measured compliance was 52.0% pre-implementation versus 62.3% post-implementation (P = 0.146); 41.8% versus 21.6% (P = 0.043) of children occluded less than 30% of prescribed occlusion time. Attendance rates in low-SES areas were 60.3% pre-implementation versus 76.0% post-implementation (P = 0.141), against 82.7% versus 84.5% elsewhere in The Netherlands. In low-SES areas, the SRIAD was 0.215 log/year pre-implementation versus 0.316 log/year post-implementation (P = 0.025); whereas elsewhere in The Netherlands, these were 0.244 versus 0.292 log/year, respectively (P = 0.005; the SRIAD's improvement was significantly better in low-SES areas than elsewhere, P = 0.0203). This advantage remained after adjustment for initial age and intraocular visual-acuity difference, cause of amblyopia, and was especially pronounced in children whose mothers spoke Dutch poorly (P = 0.022). Overall, 25.1% versus 30.1% (P = 0.038) had completed occlusion therapy after 15 months.

Conclusion. After implementation of the cartoon, electronically measured compliance improved, attendance improved, acuity increased more rapidly and treatment was shorter. This may be due, in part, to additional measures such as the course on compliance. However, the fact that these advantages were especially pronounced in children in low-SES areas with a large proportion of immigrants who spoke Dutch poorly supports its use in such areas.

INTRODUCTION

Amblyopia – a 'lazy eye' – is a reduction of visual acuity, usually unilateral, that affects approximately 3% to 4% (Attebo et al. 1998) of the general population. Until age six, amblyopia is treated with glasses and by occluding the better eye with a patch for several hours a day over a period of months or years (Awan et al. 2010). Approximately one third (Smith et al. 1995) of affected children do not attain sufficient visual acuity to read properly with the amblyopic eye (Rahi et al. 2002b). In most cases, this is due to noncompliance with occlusion therapy, that is, because the eye is not patched according to the orthoptist's prescription (Loudon et al. 2006; Smith et al. 1995). Noncompliance leads to an avoidable increase in use of healthcare resources, because it contributes to repeated office visits (see Chapter 4), missed outpatient appointments, changed treatment methods, additional prescriptions, and a longer course of illness and treatment (Lehane et al. 2009; Stewart et al. 2007a; Winnick et al. 2005).

In an earlier randomised controlled trial in The Hague (The Netherlands), we demonstrated that poor compliance with occlusion therapy was associated with low initial visual acuity, parental country of origin, mother's poor fluency in Dutch, a low level of parental education (Loudon et al. 2006), psychological distress caused by occlusion therapy (Loudon et al. 2009), and low social cohesion of parents within society (Chapter 6). This poor compliance was greatly improved by an educational cartoon that explained, without words, why children should wear the eye patch; it was accompanied by a calendar with stickers. The cartoon story uses no words because in The Netherlands, amblyopia is mostly detected at approximately four years of age because of population-based vision screening (Chapter 2) (Loudon et al. 2006). For the parents, an information leaflet in six languages (Dutch, English, French, German, Turkish and Arabic) (Appendix IX) was included with the cartoon. This was intended to supplement, not replace, the regular explanations given by the treating orthoptist. Mean electronically measured compliance in children who used the cartoon was 78%, against 56% in those who did not (Loudon et al. 2006).

In an implementation study, we assessed the degree of implementation of this cartoon among orthoptists in daily practice and the clinical effects of it among amblyopic children. The current chapter reports on its clinical effects. Because compliance was particularly low among children with non-native parents who had a low educational level and spoke Dutch poorly (Loudon et al. 2006), the study was done in low socioeconomic status (SES) suburban areas of Amsterdam, Rotterdam, The Hague and Utrecht with a large proportion of immigrants and, for comparison, in other areas in The Netherlands. Finally, we used the speed of reduction in intraocular-acuity difference (SRIAD) as a measure of the cartoon's clinical effectiveness.

METHODS

Two groups of orthoptists participated: (1) nine orthoptists working in five clinics in low-SES areas and (2) 23 orthoptists working in 15 clinics elsewhere in The Netherlands (Appendix IV).

The study entailed two periods of inclusion of children who started occlusion therapy for the first time, each approximately one year (Fig. 7.1). In the first year ("pre-implementation phase"), treating orthoptists explained amblyopia and its treatment to patients and their parents as usual. After the treating orthoptists had been instructed in a course on compliance, children received the cartoon via the orthoptist at the start of their occlusion treatment ("post-implementation phase", Fig. 7.1). The course for the treating orthoptists was about compliance in general, compliance in the treatment of amblyopia, the consequences of noncompliance for public health, communication with parents who speak Dutch poorly, intercultural communication, techniques for detecting and preventing noncompliance, and the implementation of these techniques within a hospital setting.

Patient selection

Patients aged three to six years who had been newly diagnosed with amblyopia and who had started occlusion therapy for the first time were eligible. All had an intraocular difference in visual acuity between both eyes of at least two logMAR (logarithm of the Minimum Angle of Resolution) lines after refractive correction, and all had an amblyogenic risk factor: strabismus without alternating fixation, anisometropia, or astigmatism. Children were excluded if they had undergone earlier treatment for amblyopia, mental retardation or vision loss due to an organic cause. Written informed consent by the parents or guardian was a prerequisite for participation.

In our previous randomised controlled trial (Loudon et al. 2006), the duration of occlusion (number of hours per day) for the first prescription was standardised in a focus group consisting of the study committee and treating orthoptists who participated in the study. They were given example cases of people of various ages, visual acuity and causes of amblyopia, and were asked to prescribe the number of occlusion hours per day. Cause of amblyopia proved to be of little importance, and the relationship between the two other parameters (visual acuity and age) could be represented by the following: - *6.63 x acuity of the amblyopic eye in decimals / acuity of the better eye in decimals + 0.5 x age in years + 4.97* (Loudon et al. 2006). For example, for a 3-year-old child with an acuity ratio of 0.6, the number of hours would then be: - 6.63 x 0.6 + 0.5 x 3 + 4.97 = approximately 2.5 hours of occlusion per day. It was not possible to standardise subsequent prescriptions of occlusion therapy, because treating orthoptists prescribe treatment individually according

to treatment success. All patients were examined every two to four months by their treating orthoptist.

The educational cartoon

The educational cartoon story and the calendar with stickers had been developed for the previous randomised controlled trial (Loudon et al. 2006) by two artists (J. Vingerling and G. de Bruyne) who specialized in art for sick children (Appendix IX). These elements and the parental information sheets in six languages were bound together into one booklet.

For this study, minor improvements were made on the basis of comments by parents from the previous randomised control trial, the cartoon now giving more importance to the visits to the clinic. The cartoon story was also more focussed on four-year-old children, because in The Netherlands most of the amblyopic children start with occlusion therapy at that age. This is because of the regularly performed population-based vision screening program, which has a coverage of almost 100% (Statistics Netherlands). In the Rotterdam AMblyopia Screening Effectiveness Study (RAMSES) (Chapter 2), a 7-year follow-up study of a cohort of 4,624 children born in Rotterdam in 1996/1997, it was demonstrated that most amblyopia was detected at the age of 3 years and 9 months. These children often had their first visit to the orthoptists and ophthalmologist at the age of four years. In addition, children who were recruited in our previous randomized controlled trial on the effectiveness of the cartoon were 4.5 years old, on average, when they started occlusion therapy for the first time (Loudon et al. 2006). A schedule was included at the beginning of the booklet in which the treating orthoptist could note per visit which eye needed to be patched and for how long.

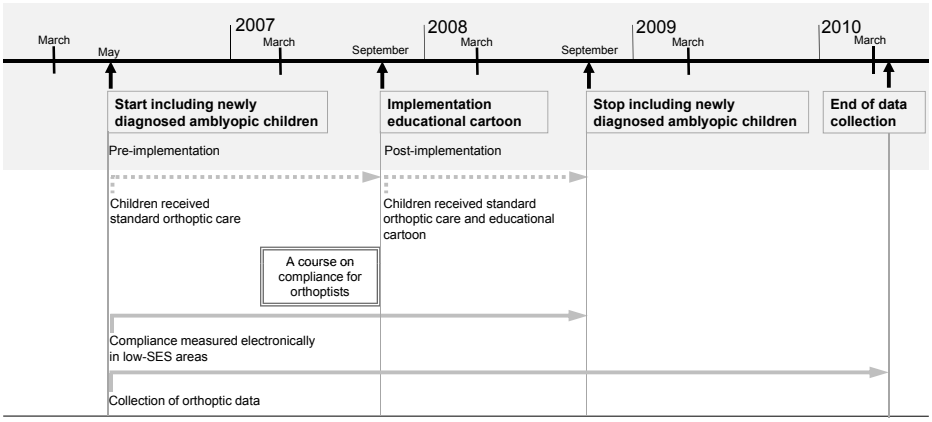


Figure 7.1) Study timetable. The events shown refer to the clinics in low-SES areas. Clinics elsewhere in The Netherlands started three months later. SES = socioeconomic status.

Data collection

After each orthoptic examination of each child or if a child missed an appointment, a standard examination form was forwarded to the research centre. This was repeated until occlusion treatment ended or until 16 months of follow-up of the last recruited child to the study.

In the low-SES areas, children's compliance with occlusion therapy was measured electronically over an entire week with an Occlusion Dose Monitor (ODM). The parents of these children were contacted by phone between the start of occlusion therapy and the second visit to the clinic. Verbal consent was obtained and an appointment for a home visit was made. During this visit, the researcher explained the use of the ODM, and full written informed consent was obtained. After its attachment to the front of the occlusion patch with double-sided adhesive tape, the ODM measured the temperature difference between the front and back of the ODM every two minutes (Chopovska et al. 2005). The researcher collected the ODM in a second home visit a week later. If a recording had failed for technical reasons or because the ODM was lost, the ODM measurement was repeated the following week. The socioeconomic and ethnic backgrounds of the families of the recruited children were assessed on the basis of a structured questionnaire (Loudon et al. 2006).

However, because the electronic measurements required at least two home visits to be made (by bicycle), all home visits to children were restricted to children living less than 5 km from the local clinics in the low-SES areas.

Mother's fluency in Dutch was rated on a five-point-scale (Loudon et al. 2006) with 1 ("not fluent") representing not speaking Dutch at all; 2 ("poor fluency") representing scarcely fluent; 3 ("moderate fluency") representing understandable and able to engage in conversation; 4 ("good") representing non-native but fluent; and 5 ("excellent fluency") representing native speaker. During the first home visit, the researcher rated the fluency in Dutch of the mother of the children included in low-SES areas in the four large cities. The fluency in Dutch of the mother of the children included elsewhere in the Netherlands was rated by the treating orthoptists, according to the same five-point-scale.

Outcome measures

The primary outcome measure of the implementation study was the degree of implementation. Two outcome measures of effectiveness are reported in this Chapter:

(1) The first outcome measure was electronically measured compliance with occlusion therapy in low-SES areas, expressed as the actual occlusion time measured with the ODM

divided by the occlusion time prescribed, this result being expressed as a percentage. In addition, average actual occlusion time measured with the ODM was assessed; this was expressed in hours of patch wearing per day. Associations were assessed between the electronically measured compliance and the following potential predictors: initial intra-ocular acuity difference, cause of amblyopia, age at start of occlusion therapy, parental country of origin, and parental fluency in Dutch. To avoid any potential influences caused by the implementation of the educational cartoon, these associations were assessed in the pre-implementation group.

(2) The second outcome measure was the speed of improvement in the intraocular visual-acuity difference, which was equal to the SRIAD. If visual acuity improved faster, one would expect the duration of treatment to be shorter. We therefore assessed the effect of the implementation on the treatment duration by determining the percentage of children that finished occlusion therapy successfully after a period of 15 months.

The charts used by the participating orthoptists to assess visual acuity of the included children depended on the age: children aged 3 to 4 years: Amsterdam Picture Chart (uncrowded, linear optotypes; Medical Workshop, Oculus, Groningen, The Netherlands); children aged 4 to 5 years: E-chart (uncrowded, linear optotypes; Medical Workshop, Oculus); children aged ≥5: Landolt-C (uncrowded; linear optotypes; Medical Workshop, Oculus). One orthoptist used the LEA Symbols (uncrowded; linear optotypes; Good-Lite, Elgin, IL) to assess the visual acuity for all age groups. Orthoptists measured acuity in decimal scores. To avoid errors in the results due to different follow-up times between the pre-implementation and post-implementation groups, we used visual-acuity data collected until end of occlusion treatment or until 16 months after start of the occlusion treatment (i.e., the shortest follow-up time of the last included child to the study). In addition, similar potential predictors (as described earlier) of the SRIAD were explored in the pre-implementation group.

Finally, we determined the outpatient attendance rate, that is, the percentage of children who had missed an appointment at least once and the percentage of children who dropped out after missing an appointment.

Statistical analysis

The severity of amblyopia was expressed as the difference in visual acuity between both eyes in log. Because of the variations in determining acuity that occur according to the age of the child and the preference of the clinics, we chose this ratio rather than the absolute acuity in the amblyopic eye.

Chi-square tests and unpaired *T*-tests were used to evaluate differences in the baseline characteristics between the pre- and post-implementation groups. The same analyses were used to examine for differences in the baseline characteristics between children living in low-SES areas of the four large cities and those living elsewhere in The Netherlands. The results for children living in the low-SES areas and for children living elsewhere in The Netherlands are presented separately.

Least-squares regression analysis and X^2 tests were used to establish differences in the electronically measured compliance and differences in the percentage of attendance rate of the pre- and post-implementation groups, and to explore predictors of the electronically measured compliance and attendance rate. These analyses were performed with SPSS 16.0.

Because the orthoptic data of each recruited child are by nature longitudinal, we used *general linear mixed models* to compare the SRIAD in the pre- and post-implementation group. We used the visual-acuity difference at the end of the observational period (15 months) to determine the rapidity of improvement between the start of the therapy and the end of the observational period or the end of treatment. Because most improvement in visual acuity is accomplished in the first weeks of treatment (Stewart et al. 2002), we tried to model this in a hyperbolic function, but uncertainty about the true nature of the relationship prevented us from going further in that regard. The linear mixed models included a categorical predictor that indicated membership of one of these two groups, a variable indicating the time elapsed between the first visit at which the child received occlusion treatment and the subsequent visits, and the interaction between these two variables. To account for the longitudinal character of the data, we included a random intercept for the child in the model. A similarly random slope of elapsed time was also included in the models. The dependent variable in this model was visual acuity difference between both eyes.

Several factors that may influence the SRIAD were investigated: the intraocular difference in visual acuity in logMAR between both eyes at start of occlusion therapy (initial intraocular visual-acuity difference), age at start of occlusion therapy, cause of amblyopia, mothers' fluency in Dutch, electronically measured compliance, mothers' country of origin, and mothers' level of education. Variables were added to the model when they had a relation with a statistical significance of $P < 0.200$.

In addition, we assessed the discrepancy between the improvement of the SRIAD after implementation in low-SES areas and the improvement of the SRIAD after implementation elsewhere in the Netherlands by use of Students-*T*-test.

All analyses regarding the SRIAD were done using SAS 9.2 (SAS Inc, Cary, NC). $P < 0.05$ indicated statistical significance.

RESULTS

In low-SES areas, 137 children were included in the study before the implementation; of these, 114 were used for analysis, six had withdrawn and 17 were not eligible. After implementation, 79 children were included the study; of these 65 were included in the analysis, six withdrew, and eight were not eligible. Elsewhere in The Netherlands, 404 children were included in the study before implementation; of these 335 were used for analysis, 24 withdrew, and 45 were not eligible. After implementation, 359 children were included in the study; of these 249 were included in the analysis, 33 withdrew, and 77 were not eligible.

Patients' demographics

Table 7.1 shows the baseline characteristics of children in low-SES areas and elsewhere in The Netherlands, and subdivided according to whether the children were in the pre- or post-implementation group. Between the groups in low-SES areas and those elsewhere in The Netherlands, there were small but significant differences in mean age at start of treatment, type of amblyopia, and mother's fluency in Dutch (Table 7.1). In low-SES areas, children were an average of five months older at start of occlusion therapy; in more cases, their amblyopia had been caused by a combined mechanism, that is, amblyopia caused by both strabismus and anisometropia. In low-SES areas, mothers' fluency in Dutch was worse.

There were no significant differences with regard to patients' demographics, mothers' fluency in Dutch, and initial intraocular visual-acuity difference between the pre- and post-implementation groups (Table 7.1). The initial intraocular visual-acuity difference was slightly larger in the post-implementation group than in the pre-implementation group: In low-SES areas it was 0.382 (Standard Deviation (SD) 0.271) log versus 0.437 (SD 0.360) log ($P = 0.246$; Table 7.1); elsewhere in The Netherlands, it was 0.406 (SD 0.286) log versus 0.437 (SD 0.331) log ($P = 0.240$; Table 7.1).

Electronic compliance measurements

Before implementation, 108 of the 114 children in low-SES areas received the ODM device for the electronic compliance measurements. Parents of the six remaining children refused to participate in the compliance measurements. Because some of the measurements failed and some of the ODM devices did not start the recording, 79 compliance

Table 7.1) Patient characteristics of children living in low-SES areas of the four large cities and elsewhere in The Netherlands subdivided according to whether the children were in the PRE-implementation or POST-implementation group. The *P*-values in the two right-hand columns show the difference in baseline characteristics between the group living in low-SES areas and the group living elsewhere in The Netherlands.

	Low-SES areas			**Elsewhere in The Netherlands**			**Difference between low-SES areas and elsewhere in The Netherlands**	
	PRE	**POST**	**Difference between PRE and POST**	**PRE**	**POST**	**Difference between PRE and POST**	**PRE**	**POST**
	n (%)	n (%)	*P*-value	n (%)	n (%)	*P*-value	*P*-value	*P*-value
Number of children included	114	65		330	252		**<0.0001***	**<0.0001***
Gender			0.279			0.315	0.588	0.164
Male	61 (53.5)	29 (44.6)		166 (50.3.0)	138 (54.8)			
Female	53 (46.5)	36 (55.4)		164 (49.7)	114 (45.2)			
Mean age at start of occlusion treatment (years)	4.6 SD 1.1	4.4 SD 1.2	0.196	4.2 SD 1.1	4.2 SD 1.1	0.320	**0.001***	**0.040***
Cause of amblyopia			0.632			0.427	**0.004***	**0.002***
Strabismus	26 (22.8)	12 (18.5)		62 (18.8)	57 (22.6)			
Anisometropia	50 (43.9)	26 (40.0)		171 (51.8)	126 (50.0)			
Combined mechanism	34 (29.8)	23 (35.4)		62 (18.8)	38 (15.1)			
Uncertain	4 (3.5)	4 (6.1)		35 (10.3)	31 (12.3)			
Mean initial intraocular visual-acuity difference	0.382 SD 0.271	0.437 SD 0.360	0.246	0.406 SD 0.286	0.437 SD 0.331	0.240	0.418	0.926
Number of prescribed occlusion time at start of occlusion treatment (hours/day)	3:37 SD 1:14	3:41 SD 1:25	0.746	3:46 SD 1:21	3:57 SD 1:15	0.090	0.437	0.157

Mother's fluency in Dutch			0.760			0.343	**<0.0001***	**<0.0001***
Excellent	46 (40.4)	26 (40.0)		296 (89.7)	234 (92.9)			
Good	26 (22.8)	19 (29.2)		17 (5.2)	10 (4.0)			
Moderate	24 (21.1)	11 (16.9)		16 (4.8)	7 (2.8)			
Poor	13 (11.4)	8 (12.3)		1 (0.3)	0 (0.0)			
None	5 (4.4)	2 (1.5)		0 (0.0)	1 (0.4)			
Mother's country of origin †			0.584				-	-
The Netherlands	39 (34.5)	22 (34.9)		-	-			
Turkey	12 (10.6)	11 (17.5)		-	-			
Morocco	26 (23.0)	12 (19.0)		-	-			
Suriname or Antilles	11 (9.9)	6 (9.5)		-	-			
Other	25 (22.1)	12 (19.1)		-	-			
Mother's highest level of education †			0.604				-	-
University	6 (5.3)	6 (9.5)		-	-			
Higher education	23 (20.3)	12 (19.1)		-	-			
Secondary education	42 (37.2)	23 (36.5)		-	-			
Primary education	32 (28.3)	14 (22.3)		-	-			
None	10 (8.9)	8 (12.7)		-	-			

SD = standard deviation; SES = socioeconomic status.

* *P*-value < 0.05.

† Data on country of origin and level of education were collected only for patients living in low-SES areas in the four large cities

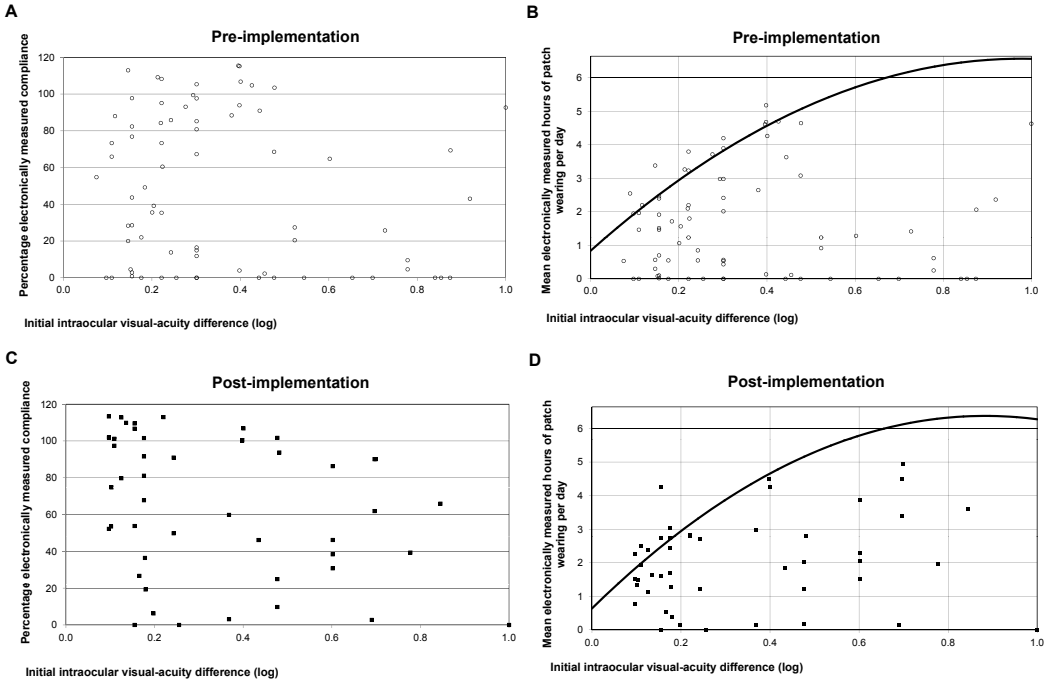


Figure 7.2) Compliance expressed as the percentage of occlusion time measured electronically divided by the prescribed occlusion time (A, C, left) and electronically measured occlusion time expressed in hours of patch wearing per day (B, D, right) against the difference in visual acuity at the start of occlusion therapy. Each symbol represents one child who received occlusion therapy for the first time. The severity of amblyopia increases towards the right side of each graph. For the first prescription of occlusion hours the number of hours was calculated according to the following formula: *-6.63 x acuity amblyopic eye / acuity fellow eye + 0.5 x age + 4.97*, reflecting average prescription behaviour among orthoptist in our previous study (Loudon 2006) (black line on right).

measurements that had been obtained before the second visit to the clinic could be used for analysis (Fig. 7.2, left). After implementation, compliance measurements regarding 51 of the 65 children could be used for analysis (Fig. 7.2, left).

Mean compliance averaged 52.0% in the pre-implementation group and 62.3% in the post-implementation group. Both had a bimodal distribution (Fig. 7.3), that is, a lot of children did not patch at all. Although mean compliance did not differ significantly between the groups (P = 0.146), 41.8% children in the pre-implementation group patched less than 30% of the hours prescribed; 19% did not even patch at all. Post-implementation, 21.6% patched less than 30% of the hours prescribed; 7.8% did not patch at all (P = 0.043).

In addition, electronically measured compliance was correlated with cause of amblyopia (P = 0.010): whereas mean electronically measured compliance was 67% in children in the pre-implementation group and in those whose amblyopia was caused by strabismus,

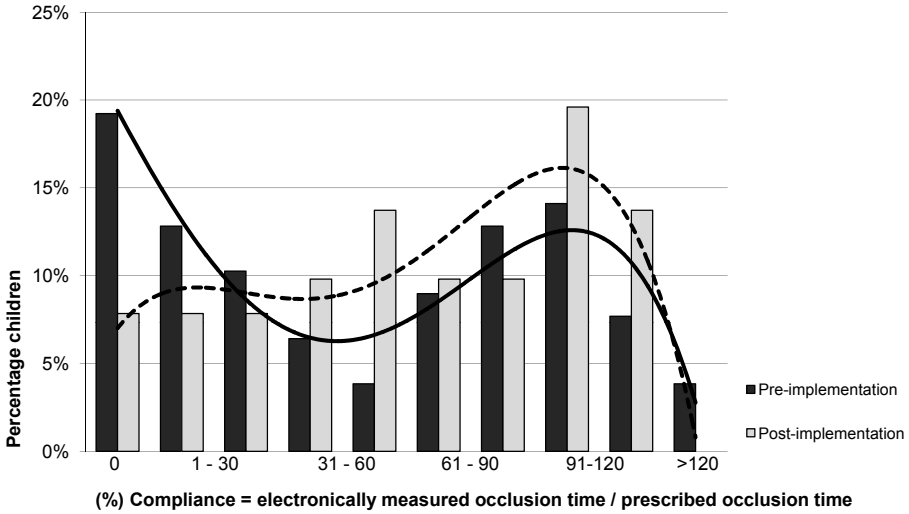


Figure 7.3) Bar chart of electronically measured compliance of the children living in low-SES areas of the four large cities (in 15% intervals). The ordinate represents the percentage children, and the abscissa is the compliance (%).Black bars: pre-implementation (n = 79). Gray bars: post-implementation (n = 51). Black line: bimodal distribution of the compliance measurements of the pre-implementation group. Dashed line: bimodal distribution of the compliance measurements of the post-implementation group.

it was 32% in those with combined-mechanism amblyopia and 41% in those with anisometropia amblyopia. Electronically measured compliance was not influenced by mother's fluency in Dutch (P = 0.504), level of education (P = 0.877) or country of origin (P = 0.212).

Figure 7.2 (right) depicts compliance expressed as hours per day of patch wearing, measured electronically, compared with the initial intraocular visual-acuity difference. Pre-implementation, most children (42.3%) occluded less than one hour; post-implementation, most children occluded between one and three hours (54.9%, P = 0.023). The mean number of hours patch wearing per day was 1:44 SD 1:35 hrs in the pre-implementation group and 2:06 SD 1:25 hrs in the post- implementation group: a difference of 21.15% in time (P = 0.176).

Attendance rate

In low-SES areas, 31 of the 78 children (39.7%) in the pre-implementation group had not attended an appointment at least once (Fig. 7.4). Twelve of these were lost to follow-up because they never visited the clinic again after missing their appointment. Post-implementation, 12 of the 50 children (24%) had not attended at least one appointment (Fig. 7.4). After they had missed their appointment, five of these children never made a new one (P = 0.182). Elsewhere in The Netherlands, 57 of the 330 children (17.3%) in the pre-implementation group missed an appointment at least once; against 15.5% (39/252) in

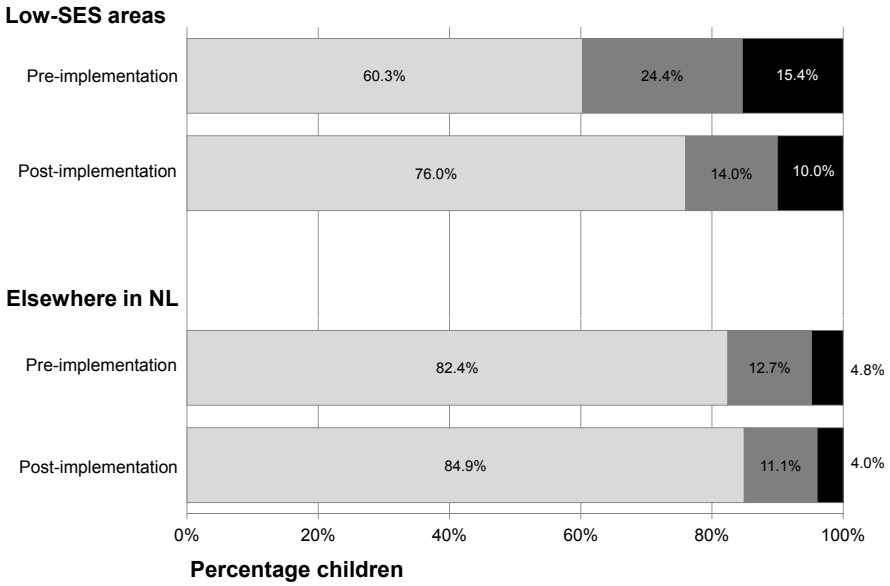


Figure 7.4) Attendance rates subdivided into low-SES areas in the four large cities and to elsewhere in The Netherlands, and subdivided according to whether children were in the pre- implementation group (n = 78 vs. n = 330, respectively) or post-implementation group (n = 50 vs. n = 252). Light-gray bars: never missed an appointment. Dark-gray bars: missed an appointment but attended next appointment. Black bars: dropped out after missing an appointment. NL = The Netherlands; SES = socioeconomic areas.

the post-implementation group. After missing their appointment, 4.8% of the children in the pre-implementation group were lost to follow-up, and 4% in the post-implementation group (P = 0.808; Fig. 7.4).

No predictors were found for attendance. However, a small trend was found that children of mothers with poor to no fluency in Dutch had a better attendance rate after implementation than before it (P = 0.177). Before implementation, 58% (n = 11) of the children of mothers with poor to no fluency in Dutch had missed an appointment at least once; that was 9% (n = 1) after implementation.

Speed of Reduction in Intraocular-Acuity Difference

To calculate the SRIAD averaged over 15 months of observation, we used visual-acuity data on 3,066 visits to the clinic of 703 of the 763 children. The mean number of visits per child was 4.36 (range 1-10). The remaining 60 children were excluded from the SRIAD analysis, as patients were lost to follow-up after the first visit, or data on mothers' fluency in Dutch, country of origin, level of education, or electronically measured compliance in low-SES areas were missing.

Table 7.2) Mean speed of reduction in intraocular-acuity difference (unadjusted and adjusted) in log over one year of occlusion treatment averaged over a 15-month observation period*

	PRE	POST	*P*-value (difference between PRE and POST)	
	Entire study population			
	n = 402	n = 301		
Unadjusted SRIAD	0.235 SD 0.199	0.296 SD 0.210	0.0004	
Adjusted SRIAD	0.238 SD 0.194	0.293 SD 0.205	0.0003†	
	Low-SES areas			**Advantage in SRIAD between PRE and POST**
	n = 78	n = 50		
Unadjusted SRIAD	0.215 SD 0.230	0.316 SD 0.262	0.0254	0.101 SD 0.243
Adjusted SRIAD	0.205 SD 0.224	0.320 SD 0.250	0.0092‡	
	Elsewhere in NL			*P*-value § = 0.0203
	n = 324	n = 251		
Unadjusted SRIAD	0.239 SD 0.193	0.293 SD 0.202	0.0046	0.054 SD 0.197
Adjusted SRIAD	0.244 SD 0.188	0.289 SD 0.197	0.0060	

SD = standard deviation; SES = socioeconomic status; SRIAD = speed of reduction in intraocular acuity difference.

* Mean SRIAD was averaged over a 15-month observation period for the entire study population (i.e., low-SES areas and elsewhere in The Netherlands), subdivided according to whether children were in the PRE- or POST-implementation group. We used visual-acuity data on 3,066 visits to the clinic for these analysis.

† Analysed with the Linear Mixed Model, which included a random patient-specific intercept, a random intercept for treating orthoptists, and a similarly random slope of elapsed time in the model, and adjusted for initial intraocular visual-acuity difference, cause of amblyopia, and mothers' fluency in Dutch.

‡ Analysed with the Linear Mixed Model, which included a random patient-specific intercept, a random intercept for treating orthoptists, and a similarly random slope of elapsed time in the model, and adjusted for initial intraocular visual-acuity difference (log) and cause of amblyopia.

§ *P*-value indicates the significant difference in the advantage in SRIAD in low-SES areas against the advantage in SRIAD elsewhere in The Netherlands. The advantage in SRIAD in low-SES was even more pronounced when we used the adjusted SRIAD rates; however, this analysis was too complex due to the multiple confounders and, therefore, this would not be acceptable to perform such a test.

Table 7.3) Mean unadjusted speed of reduction in intraocular-acuity difference (SRIAD) in log over one year of occlusion treatment averaged over a **9-month** observation period*

	PRE	**POST**	***P*-value (difference between PRE and POST)**
	Entire study population		
	n = 402	n =301	
Unadjusted SRIAD	0.456 SD 0.210	0.520 SD 0.355	0.0436
	Low-SES areas		
	n = 78	n =50	
Unadjusted SRIAD	0.351 SD 0.403	0.494 SD 0.424	0.0581
	Elsewhere in The Netherlands		
	n = 324	n =251	
Unadjusted SRIAD	0.482 SD 0.410	0.524 SD 0.407	0.2224

SD = standard deviation; SES = socioeconomic status; SRIAD = speed of reduction in intraocular acuity difference.

* Mean SRIAD was averaged over a 9-month observation period for the entire study population (i.e., low-SES areas and elsewhere in The Netherlands) subdivided according to whether children were in the PRE- or POST-implementation group. We used visual-acuity data on 2,338 visits to the clinic for these analyses.

In the entire pre-implementation group (n = 402), the mean SRIAD was 0.235 (SD 0.199) log per year. In the entire post-implementation group (n = 301), the mean SRIAD was 0.296 (SD 0.210) log per year (P = 0.0004; Table 7.2). In other words, the difference in visual acuity between both eyes decreased by 2.4 lines per year before implementation, and 3 lines per year after implementation.

In the pre-implementation group in low-SES areas (n = 78), the mean SRIAD was 0.215 (SD 0.230) log per year. In the post-implementation group in low-SES areas (n = 50), the mean SRIAD was 0.316 (SD 0.262) log per year (P = 0.0254; Table 7.2). In other words, the difference in visual acuity between both eyes decreased by 2.2 lines per year before implementation, and 3.2 lines per year after implementation.

In the pre-implementation group elsewhere in The Netherlands (n = 324), the mean SRIAD was 0.239 (SD 0.193) log per year. In the post-implementation group elsewhere in The Netherlands (n = 251), the mean SRIAD was 0.293 (SD 0.202) log per year (P = 0.0046; Table 7.2). In other words, the difference in visual acuity between both eyes decreased by 2.4 lines per year before implementation, and 2.9 lines per year after implementation.

Part of the higher SRIAD after implementation could be attributed in part to other compliance-enhancing measures, such as the course on compliance. Therefore we compared the

improvement in SRIAD in low-SES (0.316 - 0.215 = 0.101 (SD 0.243) log per year) against the improvement in SRIAD elsewhere in The Netherlands (0.293 – 0.239 = 0.054 (SD 0.197) log per year). We found that the SRIAD in low-SES improved significantly better after implementation than elsewhere in The Netherlands (P = 0.0203, *T*-test).

Averaged over a period of 9 months' observation, the SRIAD was slightly higher than depicted earlier (Table 7.3); the dissimilarity in SRIAD between pre- and post-implementation groups remained detectable. We used visual-acuity data on 2,338 visits to the clinic of 703 children for these analyses.

Univariate analysis of the entire study population included during the pre-implementation period showed several factors that were associated with the SRIAD: initial intraocular visual-acuity difference (P < 0.0001), age at start of treatment (P < 0.0001), cause of amblyopia (P = 0.0013), and mother's fluency in Dutch (P = 0.154).

The relationship between initial intraocular visual-acuity difference and age at start of treatment could be represented as: *SRIAD = – 0.01938 – 0.5669 x initial intraocular visual-acuity difference + 0.00427 x age*. This means that visual acuity increased more rapidly in younger children (despite longer prescribed patching hours in older children) and in children with severe amblyopia (partly due to longer prescribed patching hours).

The SRIAD is correlated with the type of amblyopia, with a best-to-worst ranking of 0.29 (SD 0.285) log per year for combined mechanism amblyopia, 0.26 (SD 0.188) for strabismus, and 0.23 (SD 0.188) anisometropia. This can be explained, at least in part, by the differences in initial intraocular visual-acuity difference between the causes of amblyopia. Children whose cause of amblyopia was uncertain had a mean SRIAD of 0.16 (SD 0.187) log per year.

The dissimilarity in SRIAD between pre- and post-implementation groups was especially pronounced in children whose parents spoke Dutch poorly (Fig. 7.5). Pre-implementation (n = 16), SRIAD was 0.13 (SD 0.136) log per year; post-implementation (n = 7) it was 0.39 (SD 0.227) log per year in children of mothers who spoke Dutch poorly (P = 0.022).

After linear mixed-model analysis had been used to adjust the SRIAD for these confounding factors (Table 7.2), the SRIAD in the post-implementation group remained higher than that in the pre-implementation group.

In the entire study population, we found an indication that children in the post-implementation group finished the occlusion therapy sooner than those in the pre-implementation

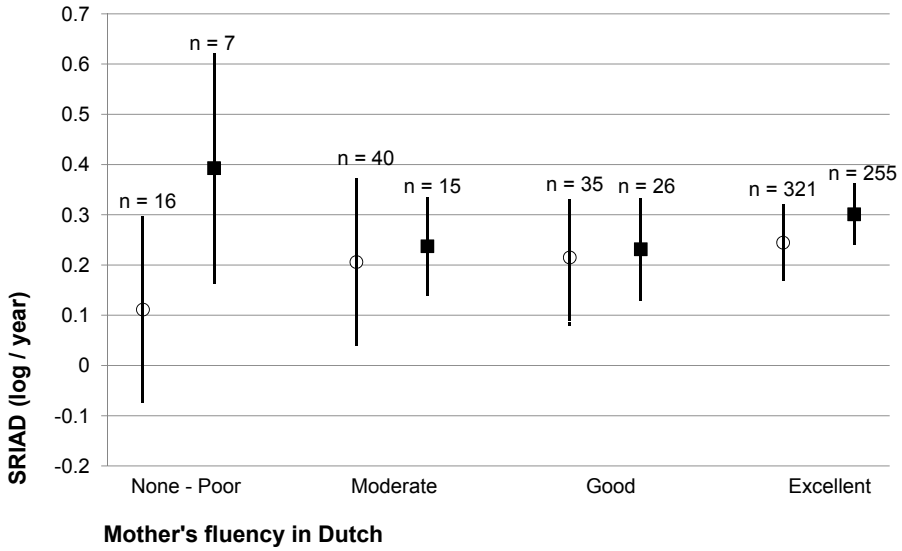


Figure 7.5) Mean SRIAD in log per year averaged over 15 months of observation, per category of mother's fluency in Dutch. Data have been corrected for visual-acuity difference at start of treatment (0.397 SD 0.180 logMAR pre-implementation and 0.342 SD 0.232 logMAR post-implementation) and age at start of treatment (4.32 SD 1.09 years pre-implementation and 4.20 SD 1,08 years post-implementation) (P = 0.091). The white circles (■) represent children in the pre-implementation group; the black squares (⑤) represent children in the post-implementation group. The advantage of the use of the cartoon on the SRIAD in the group 'None-Poor' fluency in Dutch was significant (P = 0.022). SRIAD = speed of reduction in intraocular-acuity difference.

group. Fifteen months after starting occlusion treatment, 25.1% in the pre-implementation group had finished versus 30.1% in the post-implementation group (P = 0.038, X^2 test; Table 7.4).

DISCUSSION

We found that electronically measured compliance was improved through the use of the cartoon. Mean electronically measured compliance in low-SES areas was 52% without the cartoon, and 62% with the use of the cartoon. Attendance rate improved in low-SES areas and not at all in areas elsewhere in The Netherlands. Finally, treatment duration was shorter after implementation of the cartoon, because the intraocular acuity difference decreased more rapidly.

In the entire study population, we found that the difference between acuity of the amblyopic eye and acuity in the healthy eye decreased with 2.4 lines per year, on average, in

Table 7.4) Outcome of therapy of the entire study population at 15 months after start of occlusion therapy *

	Pre-implementation group n (%)	Post-implementation group n (%)
Therapy finished	111 (25.0)	96 (30.3)
Therapy aborted	10 (2.3)	15 (4.7)
Therapy ongoing	323 (72.7)	206 (65.0)

* Therapy had been finished by more children in the post-implementation group than in the pre-implementation group: P = 0.038.

children who did not receive the cartoon against 3 lines per year, on average, in children who received the cartoon. One can argue that this improvement in SRIAD was caused, in part, by other compliance-improving measures, such as the course on compliance, or by a bias in patient selection and other effects, such as the Hawthorne effect (Mayo 1933). However, in low-SES areas, we found that the difference between acuity of the amblyopic eye and acuity in the healthy eye decreased with 2.2 lines per year, on averaged, pre-implementation compared with 3.2 lines per year, on average, post-implementation; whereas it decreased with 2.4 pre- and 2.9 post-implementation elsewhere in The Netherlands. This supports the use of the cartoon in children in low-SES areas. It seems likely that the cartoon is able to explain to a four-year-old child why he/she should patch in a situation that the parents do not understand the explanation given by the orthoptist and thus cannot motivate the child.

In our study, the severity of amblyopia was expressed as the difference in visual acuity between both eyes in log. We used the SRIAD averaged over 15 months of observations as a measure to assess the effectiveness of amblyopia treatment.

Although it is evident that visual acuity increases most in the first weeks or months of occlusion therapy (Stewart et al. 2004b), the exact relationship between occlusion time and improvement in visual acuity remains unclear. Therefore, we used a linear relationship for this analysis.

We found that 42% of all children in low-SES areas who did not use the cartoon patched less than 30% of the occlusion time prescribed (Fig. 7.3), against 22% of those who *did* use it. For comparison, in the entire city of The Hague, Loudon and colleagues (2006) found that 15% of the children who did not use the cartoon patched less than 30% of the occlusion time prescribed, against 2% of those who did use it. We found that electronically measured compliance as expressed in percentage is particularly low in children with severe amblyopia. Children with an initial intraocular visual-acuity difference of ≥0.3 log, who did not receive the cartoon, patched 1.42 (SD 0.42) hours per day, whereas children

with the cartoon patched 2.6 (SD 0.45) hours per day. This can be explained by the fact that these children were asked to patch long hours, whereas their vision during patching was low (Loudon et al. 2006; Stewart et al. 2005). In children who had been prescribed occlusion for 12 hours a day, Stewart and colleagues (Stewart et al. 2007b) found that the electronically measured occlusion time was almost similar to that in children who had been prescribed six hours of occlusion a day.

In conclusion, the linear mixed model which was used to calculate the SRIAD allowed us to correct for several confounders: initial intraocular visual-acuity difference, cause of amblyopia, age at start of occlusion therapy and mother's fluency in Dutch. As expected, the SRIAD was higher in severe amblyopia than in mild amblyopia, partly because orthoptists patched more in severely amblyopic children. The SRIAD was lower in older children, despite the fact that orthoptists patched more in older children. The relationship between age and intraocular visual acuity at start of occlusion therapy, and SRIAD for the entire study population was as follows: *SRIAD = – 0.01938 – 0.5669 x initial intraocular visual-acuity difference + 0.00427 x age*. According to this formula, an intraocular acuity difference of 3 lines at start of occlusion therapy improves with 1.7 lines per year in a four-year old child, and with 1.6 lines per year in a six-year-old child. An intraocular acuity difference of 6 lines at start of occlusion therapy improves with 3.4 lines per year in a four-year-old child, and with 3.3 lines per year in a six-year-old child.

Acknowledgments

The course on compliance for orthoptists was developed and organized in collaboration with Mrs. Marianne Sinoo of the Department of Health Orthoptics, Utrecht University of Applied Sciences. Written permission for publication of fragments of the educational cartoon in Appendix IX was obtained. Acknowledgement for proofreading and correcting the English edition of the article goes to Mr. D.R.M. Alexander of Ways with Words.

The authors thank the 20 clinics, and especially the 32 participating orthoptists, for their contributions to the study (names are listed per group, in alphabetical order of the clinic, and in alphabetical order of the names):

Amphia Hospital, Breda & Oosterhout (K. Croft, C. Klerks-Kuijpers, P. Neomagus, C. Stoop); Catharina Hospital, Eindhoven (E. van Rooijen-Troost); Flevohospital, Almere (L. Bakker, N. Bos - De Bruin); Isala Clinics, Zwolle (A. Wellner-Fokker); Kennemer Hospice, Haarlem (W. Jutte, A. Hupsch-Van den Heuvel, A. Spaan); Lievensberg Hospital, Bergen op Zoom (M. de Rooij, A. van Voorden-Van der Knaap); Máxima Medical Centre, Eindhoven & Veldhoven (E. Cockx-Huitema); Medical Clinic, Alkmaar (W. Hoogeveen, I. Huisman); Medical Centre Haaglanden, The Hague (E. van Minderhout, B. Simonsz-Tóth,

M. Vermeulen - Jongen); Mesos Medical Centre, Utrecht (A. van de Bovenkamp-Scheper, A. Versteeg); Rivierenland Hospital, Tiel (T. Kooij-'t Spijker); Ruwaard van Puttenhospital, Spijkenisse (M. Kruiswijk); Sint Lucas Andreas Hospital, Amsterdam (A. Burger-Meywaard); Tergooi Hospitals, Hilversum (D. Hamers); Twenteborghospital, Almelo (I. 't Hart-Leemhuis, M. Hazenkamp-Boekhold, F. Jansen, D. van der Nagel-Pronk); Vlietland Hospital, Vlaardingen (S. Rousse, D. Valster); and Zweeds Rode Kruis Hospital, Zierikzee, Van Weel-Bethesda Ziekenhuis, Dirksland, and Medical Centre, Middelburg (J. Kieviet-De Geus).

Chapter 8

Randomised comparison of three tools for improving compliance with occlusion therapy: an educational cartoon story, a reward calendar, and an information leaflet for parents

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Accepted for publication by Graefes Arch Clin Exp Ophthalmol. 2012; June 30

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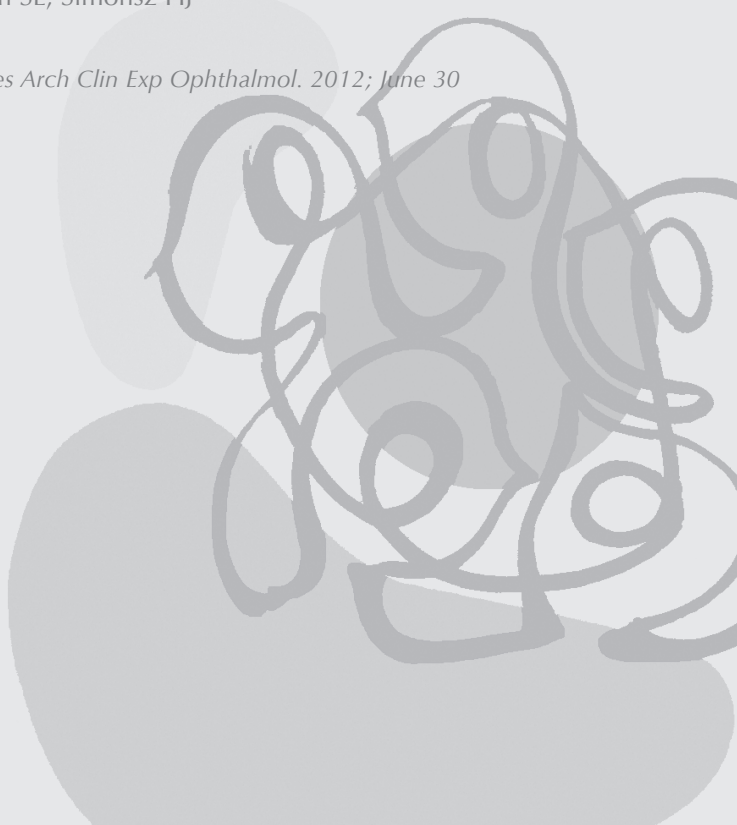

ABSTRACT

Background. We previously demonstrated that compliance with occlusion therapy for amblyopia was improved by the use of an educational programme, especially in children of parents of foreign origin and who spoke Dutch poorly. The programme consisted of (i) a cartoon story for amblyopic children that explained without words why they should patch, (ii) a calendar with reward stickers, and (iii) an information leaflet for parents. In the current study, we assessed the individual effect of each component on compliance.

Methods. We recruited 120 three-to-six-year old children who lived in a low socio-economic status (SES) area in The Hague and were starting occlusion therapy for the first time. They were randomised to receive one of the components (three intervention groups), or a picture to colour (control group). The randomisation was blinded for treating orthoptist and researcher. Compliance was measured electronically using the Occlusion Dose Monitor (ODM). Primary outcome was percentage of compliance (actual/prescribed occlusion time). Secondary outcome was absolute occlusion hours per day. Parental fluency in Dutch was rated on a five-point scale.

Results. Compliance could be measured electronically in 88 of the 120 children; in 32 others, it failed for various reasons. Parental fluency in Dutch was moderate or worse in 36.4% (P = 0.327). Average compliance was 55% Standard Deviation (SD) 40 (n = 18) in the control group, 89% SD 25 in the group receiving the educational cartoon (n = 25, P = 0.002 compared with control group), 67% SD 33 (n = 24, P = 0.301) in the reward-calendar group and 73% SD 40 (n = 21, P = 0.119) in the parent-information-leaflet group. On average, children in the control group occluded 1:46 SD 1:19 hours/day, 2:33 SD 1:18 hours/day in the group receiving the educational cartoon, 1:59 SD 1:13 hours/day in the reward-calendar group and 2:18 SD 1:13 hours/day in the parent-information-leaflet group. No child who received the cartoon story occluded less than one hour per day, against seven in the reward-calendar group, five in the parent-information-leaflet group and five in the control group.

Conclusions. Although all three components of the programme improved compliance with occlusion therapy in children in low-SES areas, the educational cartoon had the strongest effect, as it explained without words to a four-to-five year old child why it should wear the eye patch.

INTRODUCTION

With a prevalence of 3-4% amblyopia is the most common visual defect in young children (Noorden von et al. 2002; Searle et al. 2002). Occurring in the absence of an eye defect, it is a reduction in visual acuity, usually unilateral, caused by visual deprivation, or otherwise either by strabismus, a refractive error, or a combination of the two (Noorden von et al. 2002; Repka et al. 2003). The commonest method of treatment is by occluding the better eye for several hours per day (Bacal 2004; Doshi et al. 2007; Webber et al. 2005).

Various studies have shown that compliance rates for occlusion therapy are low (Awan et al. 2005; Loudon et al. 2006; Newsham 2000). As proper compliance depends greatly on good communication between patient and caregiver (Friedman et al. 2008; Tates et al. 2001; Winnick et al. 2005), communication is essential for good treatment outcome (Awan et al. 2005; Matsui 2007; Simons et al. 1999; Stewart et al. 2005; Stewart et al. 2004b). Loudon et al. (2006) found four main predictors for low compliance: low fluency in the national language, low level of education, and country of origin of the parents, and a low visual acuity of the amblyopic eye at start of occlusion therapy of the child.

Several studies have investigated ways of improving compliance. Indinnimeo et al. (Indinnimeo et al. 2009) showed an association between improved parental knowledge about asthma and a reduced number of asthma attacks in children receiving an educational programme. Holzheimer et al. (Holzheimer et al. 1998) showed that compliance with the prescribed asthma medication was better in children with asthma who had received an educational videotape, an educational book, or both.

In ophthalmology, studies on improving compliance have focussed mainly on adult glaucoma patients (Friedman 2009; Friedman et al. 2008; Friedman et al. 2009a; Okeke et al. 2009a, b). Those on improving compliance in children are scarcer. With regard to improving compliance with occlusion therapy, positive result has been reported by giving written information to the parents (Goransson et al. 1998; Newsham 2002). However, Loudon et al. (2006) found that compliance with occlusion therapy for amblyopia was significantly increased by an educational programme for children living in The Hague, which increased compliance from 57% Standard Deviation (SD) 40% to 78% SD32% (P<0.0001).

This programme was designed by José Vingerling and Gerard de Bruyne, and consisted of three components: (i) an educational cartoon story without words that explained to children why occluding was needed, (ii) a calendar with reward stickers, and (iii) an information leaflet for the parents that contained additional information on amblyopia. The cartoon story was self-explanatory, and targeted four-year-old children, four being

the age at which most children start occlusion therapy. It was designed in such a way that children from different cultures could identify with the child in the cartoon. The calendar with reward stickers was used to stimulate and motivate them: each day a child had occluded according prescription, he or she was allowed to put a sticker on the calendar. The information leaflet for the parents contained practical information about the diagnosis and therapy, and had been translated into six languages.

Although the effectiveness of the educational programme was already proven, it was not known which component of this programme played the greatest part in the increase in compliance, or whether the increase was due to the combination of the three components (Loudon et al. 2006; Okeke et al. 2009b). We therefore assessed the individual effects of each component of Vingerling and De Bruyne's educational programme, investigating among children who lived in a low socio-economic status (SES) area in The Hague.

METHODS

Patient recruitment

From October 2008 to March 2010, three orthoptists working at Haaglanden Medical Centre in The Hague recruited three-to-six-year-old amblyopic patients who were starting occlusion therapy for the first time. This hospital was situated in a low-SES area with a high proportion of non-native inhabitants. Eligible for the study were three-to-six-year old children with an intraocular difference in visual acuity between both eyes of at least two logMAR lines (i.e. logarithm of the Minimum Angle of Resolution) after refractive correction, and all had an amblyogenic risk factor: strabismus without alternating fixation, anisometropia, or astigmatism. Children were excluded if they had undergone earlier treatment for amblyopia, had a mental retardation, or had vision loss due to an organic cause.

All children were offered standard orthoptic care by of their treating orthoptist and all received a routine orthoptic and ophthalmic examination. To standardize the duration of occlusion for the first prescription, we used the same formula as in our previous study (Loudon et al. 2006). The orthoptists who participated in that study patched, on average, in relation to initial visual acuity and age: - *6.63 x acuity of the amblyopic eye in decimals / the acuity of the better eye in decimals + 0.5 x age in years + 4.97*. This was found as their average prescribing behaviour in a focus group.

Table 8.1) Five-point scale reflecting parents' fluency in Dutch (Loudon 2006)

Score	Description	Definition
1	None	Conversation with a parent could be accomplished only through an interpreter
2	Poor	Parent gave only superficial responses to the questions, using the same standard phrases and sentences
3	Moderate	Parent was unable to formulate proper Dutch sentences
4	Good	Non-native parent spoke fluent Dutch, but obviously not as his/her native language
5	Excellent	Native speaker of Dutch

Randomisation

After the treating orthoptist had obtained verbal informed consent from the parents, they randomly gave each eligible child the colouring pictures (control) or one of the three components of the educational programme. To mask this randomisation from the treating orthoptist and researchers, each component was placed in an identical-looking envelope, all of which were made the same weight. Orthoptists informed the parents that the envelope either contained one of the three components, or the colouring pictures. Parents were instructed not to tell the orthoptists or the researcher which of the four items they received in the envelope. Each envelope contained a sealed document which the parent had to give the researcher during the first home visit, and which contained information on which element the patient had received. The researcher opened this sealed document after all data had been collected. In total, 30 envelopes per component were made in advance.

Written informed consent by the parents was a prerequisite for participation, and was obtained during a subsequent house visit by the researcher.

Data collection

After the child had been to the hospital, parents were contacted by telephone and an appointment was made for a home visit. During this visit, parents were informed about the use of the Occlusion Dose Monitor (ODM), a small device that measures compliance electronically. They were asked to tape the ODM to the outside of the patch with double-sided tape every time the child was going to wear the patch. This method of compliance measurement has been proved to be reliable (Chopovska et al. 2005; Fronius et al. 2006). Compliance was measured over a one-week period and was finished before the child's second visit to the orthoptist. After this week of measurements, researchers collected the ODM. If recording by the ODM had failed due to battery failure, the parents were asked to use it for a second week.

Each mother's fluency in Dutch was rated on a five-point-scale (Table 8.1) (Loudon et al. 2006): 1 ("not fluent") meant they did not speak Dutch at all; 2 ("poor fluency") that they

were scarcely fluent; 3 ("moderate fluency") that they were understandable and able to engage in conversation; 4 ("good") that they were non-native but fluent; and 5 ("excellent fluency") that they were native speakers. Rating was done by the same researcher for all the mothers.

The socioeconomic and ethnic backgrounds of the families of the recruited children were assessed on the basis of a structured questionnaire (Loudon et al. 2006).

Outcome measures

Primary outcome measure was the electronically measured compliance, which was expressed as the actual occlusion time measured by the ODM, divided by the occlusion time prescribed by the orthoptist. Secondary outcome was the actual number of occlusion hours per day.

Statistical methods

X^2 tests and *ANOVA* were used to test for unequal distributions of patients' demographic characteristics between the four groups. The compliance rates of the three intervention groups were compared with those of the control group by using *UNIANOVA* and *Regression analysis* (SPSS 16.0). These tests were also corrected for the confounders found in the previous study (Loudon et al. 2006).

Decimal values of visual acuity were converted to logMAR. To minimize the influence of visual acuity measurement conditions, we used the logMAR difference between the visual acuities of both eyes (visual acuity difference).

The charts used by the participating orthoptists to assess the visual acuity of the recruited children depended on the child's age. The Amsterdam Picture Chart (uncrowded, linear optotypes; Medical Workshop, Oculus, Groningen, The Netherlands) was used with children aged 3 to 4; the E-chart (uncrowded, linear optotypes; Medical Workshop, Oculus) with those aged 4 to 5; and the Landolt-C (uncrowded; linear optotypes; Medical Workshop, Oculus) with those aged 5 and older.

While visual acuity was measured in decimal scores, the Amsterdam Picture Chart denotes visual acuity as a fraction, where 5/5 is considered to be equivalent to 0 logMAR. According to this chart, an intraocular difference of 2 lines indicates amblyopia, and the child should therefore start occlusion therapy. However, these fractions were hard to compute into logMAR, as there is no standardized method to compute them. We tried to compute this by calculating the fraction into decimal score and then into logMAR. As a result, some children had an intraocular difference smaller than 2 logMAR lines, even

though the chart qualified them as amblyopic (meaning that they should start patching). We did not exclude such children from the analysis.

RESULTS

Sample characteristics

We recruited 120 children to the study (Fig. 8.1), but one was excluded due to albinism. The parents of five children could not be contacted despite repeated attempts to contact them by phone (with a maximum of five attempts) and by home visit (with a maximum

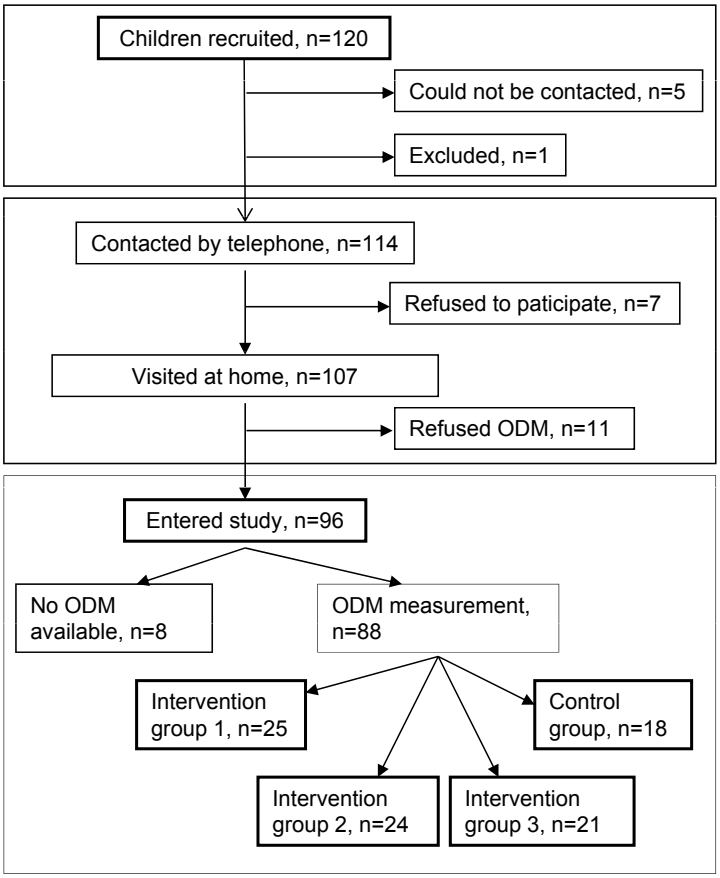

Figure 8.1) Flowchart of all children recruited. Electronic measurement of compliance was possible in 88 of the 120 children recruited. Intervention group (1) received the educational cartoon story. Intervention group (2) received the calendar with reward stickers. Intervention group (3) received the parent information leaflet in six languages. The control group received pictures to colour.

Table 8.2) Table with characteristics of the 88 children included in this trial. VA = visual acuity.

	Educational cartoon	Reward calendar	Parent information leaflet	Control group	
	n	n	N	n	P-value
Number of children included	25	24	21	18	
Gender					0.916
Male	15	12	12	10	
Female	10	12	9	8	
Mean age (yrs) at start of occlusion therapy in categories					
<4	11	10	5	5	
4-5	6	7	5	8	
>5	8	7	11	5	
Mean age (yrs) at start of occlusion therapy	4.3 SD 1.0	4.4 SD 1.2	4.9 SD 1.1	4.5 SD 1.1	0.333
Cause of amblyopia					0.277
Strabismus	10	9	4	4	
Anisometropia	6	10	11	8	
Combined	7	3	6	2	
Unknown	2	2	0	4	
Initial VA difference (logMAR)					0,918
0-<0.25	14	14	9	8	
0.25-<0.5	9	10	9	9	
0.5-<0.75	0	0	2	0	
0.75-<1.0	2	0	1	1	
Mean initial VA difference (logMAR)	0.28 SD 0.20	0.24 SD 0.10	0.31 SD 0.20	0.29 SD 0.15	
Prescribed number of occlusion hours per day	2:54 SD 1:09	2:57 SD 1:02	3:27 SD 1:19	3:20 SD 0:59	0.249
Parental fluency in Dutch (mother)					0.327
Excellent	13	8	10	7	
Good	2	7	3	6	
Moderate	9	7	7	3	
Poor	1	2	0	2	
None	0	0	1	0	
Country of origin (mother)					0.944
Native Dutch	10	7	6	5	
Turkey	2	3	3	2	
Morocco	4	3	6	4	
Surinam	3	3	1	1	
Other	6	8	5	5	
Unknown	0	0	0	1	

Highest level of education (mother)					0.438
University	3	1	3	1	
Higher education	5	5	4	5	
Secondary education	10	11	7	7	
Primary education	5	7	6	4	
None	2	0	1	0	

of three attempts). Of the remaining 114 included children, the parents of 18 children withdrew from the study. One of these 24 children who did not participate the study had received the educational cartoon story, four had received the calendar with reward stickers, nine had received the parent information leaflet, and ten had received the colouring pictures.

In the remaining 96 children, compliance was measured with the ODM. No compliance data were available for eight of these 96 children. In six of these eight cases, the ODM had failed, and parents had refused to perform a second measurement period in the subsequent week. Two ODM devices were lost. Due to failure of the device in the first period, 17 of the remaining 88 children whose ODM measurements were eventually successful were asked to use the ODM a second time. To complete the data, one of these children required a third measurement. The compliance data measured ranged from 2-8 days, with an average of 5.6 days per child. All measurements were performed and completed before the next appointment with their treating orthoptist.

Twenty-five children received the educational cartoon story, 24 received the calendar with reward stickers, 21 received the parent information leaflet, and 18 received the colouring pictures (Fig. 8.1).

Patients' demographics

The baseline characteristics were similar between groups: at start of occlusion treatment, mean age was not significantly different (P = 0.333) (Table 8.2); mean intraocular acuity was 0.28 log in the cartoon-story group, 0.24 log in the reward-calendar group, 0.31 log in the parent–information-leaflet group, and 0.29 in the control group (P = 0.641).

Forty percent of parents in the cartoon-story group spoke Dutch moderately or worse, against 33% in the reward-calendar group, 38% in the parent-information-leaflet group, and 28% in the control group (P = 0.327) (Table 8.2).

Study outcome

Mean electronically measured compliance was better in each of the three intervention groups than in the control group. Mean compliance in the control group was 55.4%; the SD was 40 (Table 8.3), similar to that in the previous study (57% SD 40, n = 155, P = 0.872, exact *T*-test) (Loudon et al. 2006). Mean compliance of children who received the educational cartoon story was 88.9% SD 25 (P = 0.002, in comparison with the control group), it was 66.7% SD 33 for that of children who received the calendar with reward stickers (P = 0.301), and it was 72.9% SD 40 for that of children who received the parent information leaflet (P = 0.119).

The compliance rates of the intervention groups differed significantly between the cartoon-story and the reward-calendar groups (P = 0.011), but did not differ significantly between the cartoon-story and the information-leaflet groups (P = 0.106), and between the information-leaflet and the reward-calendar groups (P = 0.577).

Average compliance in children of parents with excellent or good fluency in Dutch was 78% SD 37, against 63% SD 33 in children of parents with moderate or poor fluency. In children with Dutch-native parents, compliance averaged 75% SD 40, against 70% SD 45 in children with parents from Surinam, 68% SD 35 in children with parents from Morocco, and 65% SD 27 in children with parents from Turkey. Average compliance was 68% SD 34 in children of parents with secondary education or lower, against 80% SD 40 in children of parents with a higher education. However, none of these differences were statistically significant.

The right-hand panels in Figure 8.2 show the relationship between the number of hours of electronically measured occlusion per day (expressed as hours per day of patch wearing) and the initial intraocular visual acuity difference. The black line in these graphs represents the first prescription of occlusion hours. Each symbol in these graphs represents one child, with the severity of amblyopia increasing towards the right in each graph. The nearer the symbol is positioned to the black line, the more compliant this child was with the orthoptist's prescription.

On average, while children who received the educational cartoon occluded for more actual hours per day (2:33 SD 1:18 hours / day), than those in the other groups, they also had the lowest average number of prescribed occlusion hours per day (2:54 SD 1:09 hours / day, Table 8.3). Mean duration vs. prescribed hours of occlusion per day were 2:18 SD 1:13 hours vs. 3:27 SD 1:19 hours in the group who received the parent information leaflet; 1:59 SD 1:13 hours vs. 2:57 SD 1:02 hours in the group who received the reward

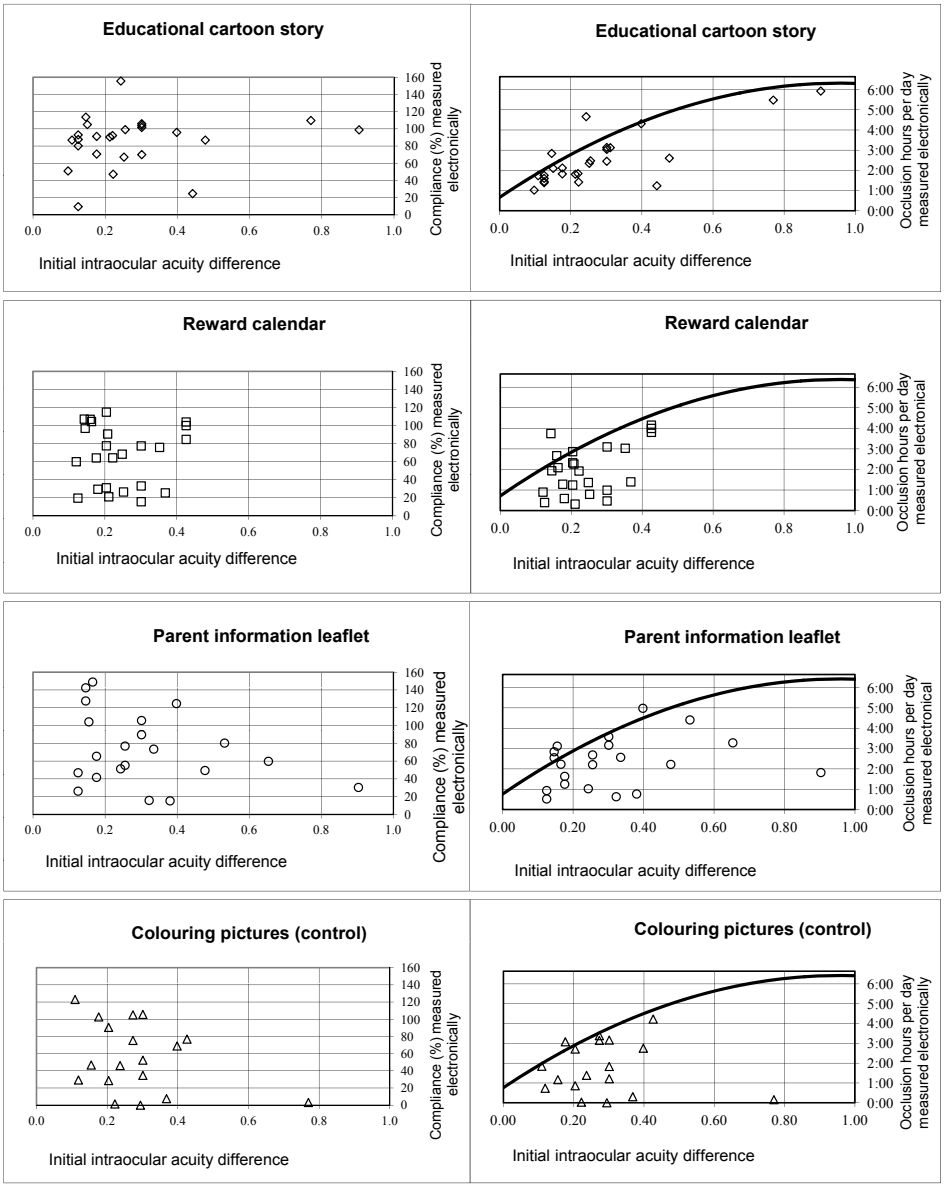

Figure 8.2) Compliance expressed as the percentage of occlusion time divided by the prescribed occlusion time (left panel) and electronically measured occlusion time expressed in hours (right panel) relative to the difference in visual acuity at the start of occlusion therapy. Each symbol represents one child who received occlusion therapy for the first time, with the severity of amblyopia increasing towards the right. For the first prescription, the following formula was used to calculate the number of hours of occlusion: *-6.63 x acuity amblyopic eye / acuity fellow eye + 0.5 x age + 4.97* (Loudon 2006). This represented the average hours prescribed by orthoptists (black line in right panels).

Table 8.3) Mean percentage of electronically measured compliance, the prescribed number of occlusion hours and the electronically measured number of occlusion hours per group.

	Mean % compliance	Prescribed number of occlusion hours/day	Electronic measured number of occlusion hours/day
Educational cartoon story (n = 25)	89 SD 25	2:54 SD 1:09	2:33 SD 1:18
Reward calendar (n = 24)	67 SD 33	2:57 SD 1:02	1:59 SD 1:13
Parent information leaflet (n = 21)	73 SD 40	3:27 SD 1:19	2:18 SD 1:13
Control group (n = 18)	55 SD 40	3:20 SD 0:59	1:46 SD 1:19

calendar, and 1:46 SD 1:19 hours vs. 3:20 SD 0:59 hours in the group who received the pictures to colour.

Of the 18 children in the control group, seven occluded less than one hour per day, against seven of the 24 children in the reward-calendar group, and five of the 21 in the information-leaflet group. No child in the cartoon-story group occluded less than one hour per day.

DISCUSSION

In children in low-SES areas whose parents spoke Dutch moderate or worse, the greatest improvement in compliance with occlusion therapy was brought by the educational cartoon, not by the calendar with reward stickers or by the parent-information leaflet. Average electronically measured compliance for children using the cartoon was significantly better than for the control group (88.9% against 55.4%, P = 0.002), despite the fact that some of the children who received the cartoon story had a large intraocular visual acuity difference at start of treatment. There were no significant differences between the control group and the groups using the parent information leaflet, or between the control group and group using reward calendar.

We have strong indications that compliance improved most in the group that received the cartoon story. In eight of the 25 children who received the cartoon story, compliance was close to 100%, against four of the 24 in the reward-calendar group, one of the 21 in the parent-information-leaflet group, and one of the 18 in the control group. Children who received the cartoon story and were measured with the ODM also patched the highest number of hours per day: unlike those in the other groups, all of them patched at least one hour per day.

Remarkably, the compliance rate in children who received only the educational cartoon story (89% SD 25, n = 25) seemed to be higher than that in children who received all three components of the educational programme (78% SD 32, n = 155) (Loudon et al. 2006). However, due to the great differences in the numbers of children, these compliance rates were not statistically different (P = 0.106, exact *T*-test).

The main limitation was the power of this study. If we had included a higher number of children, the statistical outcomes would have been different. However, as the effectiveness of the educational programme was already proven, we wished to assess the compliance rates of each component on a small scale. We therefore decided to limit our maximum inclusions to 120 children, believing this would be enough to indicate which of the three components most influenced on compliance.

With regard to the electronic compliance measurements, it was obtained for only one week, as previous studies (Chopovska et al. 2005; Loudon et al. 2006) showed that compliance measurements obtained over only one week in the first three months of the occlusion therapy best represented actual compliance. Loudon et al. (2006) found that compliance decreased during treatment, but this was due partly to a selection bias: children with low initial visual acuity were less compliant, therefore wore the patches for a longer period, and were consequently recorded more often. For this reason we decided not to measure compliance electronically after the second visit to the orthoptist.

There were fewer children in the control group than in the other three intervention groups. Ten of the 30 children who received colouring pictures did not participate in the study, possibly because their parents refused, or because they were disappointed when, on opening the envelope, they found only the colouring pictures. It may also be relevant that most parents who refused participation stated that they had no time for the home visit.

As this study recruited children living in a low-SES area with a large proportion of non-native inhabitants who spoke Dutch poorly, it is hard to indicate the value of the educational program for children living in parts of The Netherlands where such linguistic problems are less common. As the cartoon story uses no words to explain why children would wear an eye patch, it seems more likely that its effect would be greater in such low-SES areas.

Several studies have shown that the transfer of information to a child and its parents is an important factor in improving compliance in children (Friedman et al. 2008; Holzheimer et al. 1998; Indinnimeo et al. 2009; Loudon et al. 2006; Newsham 2000, 2002; Tates et al. 2001; Winnick et al. 2005). One study showed that compliance was improved more by informing both child and the parent than solely by rewarding the child (Oto et al.

2002). However, in our study, we were unable to find a significant difference in mean compliance between the group using the cartoon story and the group using the parental-information leaflet.

In conclusion, while our data shows only that compliance improved significantly in the children who used the self-explanatory cartoon story, but not in control group, we believe that a similarly designed educational cartoon story could also be useful in the long-term treatment of other diseases in young children, for example asthma being one obvious example.

Acknowledgements

The Occlusion Dose Monitor was developed in 1996-1997 as a public domain project at the department of Medical Technical Development at Amsterdam Academic Medical Centre, The Netherlands. Acknowledgement for proofreading and correcting the English edition goes to Mr. D.R.M. Alexander of Ways with Words.

Chapter 9

Implementation of an educational cartoon and other compliance-enhancing measures by orthoptists in the treatment of amblyopia

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Under review

ABSTRACT

Purpose. In an implementation study, we evaluated orthoptists' use of an educational cartoon that improves compliance with occlusion therapy for amblyopia.

Methods. Participating orthoptists provided standard orthoptic care for one year. In a second year, they supplied the cartoon in addition. They attended a course on compliance. Additional compliance-enhancing measures were planned. Orthoptists' awareness, attitude and activity in dealing with noncompliance were assessed by interviews, questionnaires and observations, and their use of the cartoon was measured. The study was performed in low socio-economic status (SES) areas and, for comparison, in other areas in The Netherlands. National implementation of the cartoon and integration of education on compliance into basic and continuing orthoptic training were attempted.

Results. Nine orthoptists in low-SES areas participated and 23 in other areas. The cartoon was used by all in low-SES areas, and by two-thirds of the others. A change in awareness and attitude was noted before and after the course on compliance, but was not sustained. Although orthoptists estimated compliance around 70% in the questionnaires, only a quarter suspected noncompliance in one patient during one day of observation. Children received a longer explanation in the second year. Hardly any of the additional compliance-enhancing measures were implemented. Integration of education on compliance into the training for orthoptists failed. The cartoon is distributed nationwide via the university clinic's valorisation centre.

Conclusion. The educational cartoon was best used in low-SES areas. Additional compliance-enhancing measures were not implemented, although these had been conceived and planned with the best intentions.

INTRODUCTION

Amblyopia (a 'lazy eye') is a decrease in best-corrected visual acuity, usually unilaterally, for which no organic cause can be found. Its prevalence is approximately 3-4% (Chapter 2) (Attebo et al. 1998). It is treated by occluding the better eye with a patch for several hours a day, and also with glasses (Holmes et al. 2006a). Approximately one-third of affected children do not attain sufficient visual acuity in the amblyopic eye (Jensen et al. 1986), due mainly to noncompliance with occlusion therapy – in other words, because the eye is not patched according to the orthoptist's prescription (Loudon et al. 2006; Smith et al. 1995). Because noncompliance contributes to repeated office visits (Chapter 4), missed outpatient appointments, changed treatment methods, additional prescriptions and a longer course of illness and treatment, it leads to the unnecessary use of healthcare resources (Chapter 4) (Lehane et al. 2009; Stewart et al. 2007a; Winnick et al. 2005). In a study in The Hague (The Netherlands (NL)), Loudon and colleagues (2006) previously demonstrated that poor compliance with occlusion therapy was correlated not only with low initial visual acuity, but also with country of origin, poor fluency in Dutch, a low level of education, psychological distress caused by occlusion therapy (Loudon et al. 2009), and low social cohesion within the society of the parents (Chapter 6). This poor compliance was greatly improved by an educational cartoon which consisted of a cartoon story that explained without words why children should wear the eye patch. The cartoon was accompanied by a reward calendar with stickers and by a six-language information leaflet for parents. The cartoon story and the calendar with stickers had originally been developed for the previous randomized controlled trial (Loudon et al. 2006) by two artists, J. Vingerling and G. de Bruyne, who specialize in art for sick children (Appendix IX).

However, it remained uncertain whether this very effective cartoon could be adopted in daily orthoptic practice. As various studies have shown, the adoption of useful research findings in day-to-day practice tends to be slow and difficult (Glasgow et al. 2003; Grimshaw et al. 2004; Grimshaw et al. 2002; Grol et al. 1999; Horner et al. 2004; Stetler et al. 2008), usually due to a range of factors – lack of agreement, lack of positive expectations with regard to the results of the new product, lack of motivation, and lack of the time or resources necessary to changing daily routines and to using this new product in daily practice (Cabana et al. 1999; Grol et al. 2005).

In this implementation study, we therefore assessed whether orthoptists were willing to adopt the cartoon. As compliance is a more common problem in suburban, non-native, low-socioeconomic status (SES) areas, where a large proportion of the population speaks Dutch poorly, we also assessed whether orthoptists in these areas would use the cartoon more often than their colleagues elsewhere in NL. Thirdly, we also assessed whether the cartoon could be made available throughout NL, and, finally, whether education on compliance could be integrated into the basic and continuing training for orthoptists. .

METHODS

Subjects & Study design

Two groups of Dutch orthoptists participated: (1) orthoptists working in clinics in non-native, suburban, low-SES areas of Amsterdam, Rotterdam, The Hague and Utrecht; and (2) orthoptists working in clinics elsewhere in NL Orthoptists working elsewhere in the NL were recruited through an appeal during a meeting of the *Nederlandse Vereniging van Orthoptisten* (NVvO), i.e., the National Orthoptic Association of The Netherlands.

The study entailed two periods, each of approximately one year (Fig. 9.1). In the first year (pre-implementation), treating orthoptists took their usual approaches to explaining amblyopia and its treatment to patients and their parents. In the second year (post-implementation), children received the cartoon from their orthoptist at the start of occlusion treatment.

The study centre collected and analysed data of the implementation. The orthoptists were asked to report to the study centre each three-to-six-year old child who had been newly diagnosed with amblyopia and who had been prescribed occlusion therapy for the first

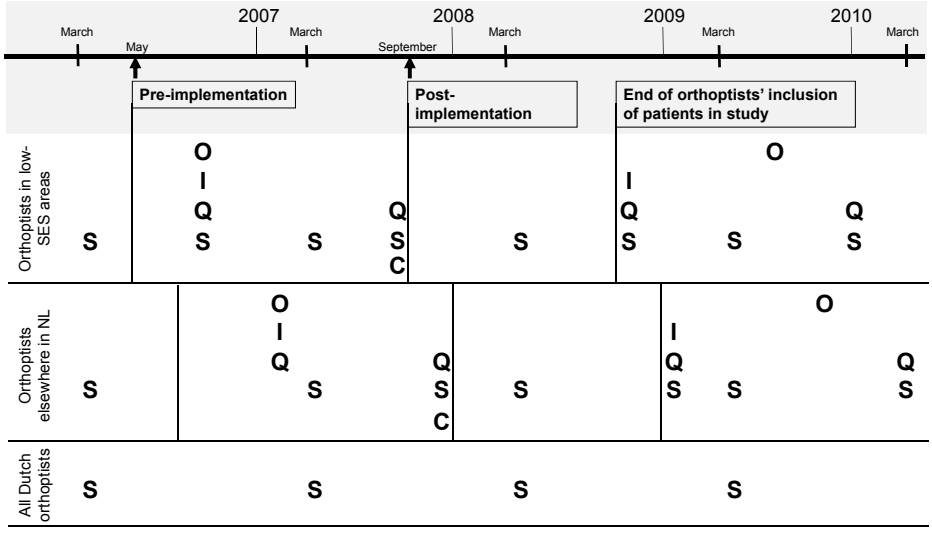

Figure 9.1) Timeline showing data collection among orthoptists in low-SES areas, orthoptists elsewhere in NL, and all Dutch orthoptists (i.e., members of the Dutch Orthoptic association (NVvO) and all orthoptists participating in the study).
O = observations in orthoptists' practices; I = semi-structured interview; Q = structured questionnaire; C = course on compliance, and S = national compliance survey

time. Biannual study-committee meetings were held to conceive and design implementation strategies. For one part of these strategies, additional compliance-enhancing measures were conceived during these study-committee meetings and introduced during the course on compliance which was offered for the participating orthoptists.

The educational cartoon

The educational cartoon story and the calendar with stickers had originally been developed for the previous randomized controlled trial (Loudon et al. 2006) by two artists, J. Vingerling and G. de Bruyne, who specialize in art for sick children (Appendix IX). These components and the parental information sheets in six languages were bound into a single booklet.

On the basis of comments by parents from the previous randomized control trial, four minor improvements were made for this study: (1) the cartoon was adapted to give greater importance to the visits to the clinic, (2) the cartoon story was more focused on four-year-old children, the age at which most amblyopic children in The Netherlands start with occlusion therapy; (3) the beginning of the booklet now included a schedule in which the treating orthoptist could note per visit which eye needed to be patched and for how long; and (4), an additional cartoon was made that explained to children why they should wear their glasses (Appendix X).

Implementation strategies

Biannual meetings were organized for all participating orthoptists, not only to promote awareness of compliance-issues, but also to gather information on the compliance issues they faced and to learn which issues they experienced in daily practice. The meetings involved discussions on the following: the inclusion of patients in the study, dealing with noncompliance with occlusion therapy, dealing with patients who do not show on appointments, and dealing with non-native patients who were not fluent in Dutch. We also kept the participating orthoptists informed on the progress of the study, asked for their feedback on the implementation process, and discussed the suitability of the educational cartoon.

Before the implementation of the cartoon, we held an accredited course on compliance (Fig. 9.1). This had three aims: to stimulate the participating orthoptists' interest in compliance issues, to create positive intentions towards handling compliance-issues, and to promote the benefit of the use of the cartoon. The course comprised interactive lectures on five themes: compliance in research and healthcare practice, the impact noncompliant behaviour at public health level, compliance with amblyopia treatment, communication techniques, and intercultural communication. The lecturers who presented the first part

of this course were all active in compliance research. These activities were followed by communication-skills training sessions in which the participants performed role-playing exercises. Each part of the course ended with a short written appraisal, and the course as a whole ended in an interactive debate between participating orthoptists and the study committee on nine pre-determined themes: (1) the use of pictorial representations to transfer information to children, (2) cost-effectiveness in the healthcare services, (3) who is responsible for successful treatment, (4) explaining amblyopia to patients with poor Dutch fluency, (5) dealing with language barriers, (6) the financial consequences of noncompliance, (7) compliance and long outpatient waiting periods, (8) unsuccessful referral after a positive vision-screening test, and (9) noncompliance in attending appointments.

On the basis of these debates and the study-committee meetings, the study committee and the participating orthoptists formulated seven additional measures to deal with noncompliance: (1) registering and calling patients who had missed an appointment, (2) registering and requesting feedback on children who had been referred for a positive vision screening test at the child healthcare centres, (3) sending a reminder to patients about the appointment, (4) organizing information sessions for parents of children who need to patch for the first time, (5) sending a letter or a bill, (6) using more often interpreters with parents who speak Dutch poorly, and (7) planning extra allotted time for a patients' first visit.

To evaluate which of these additional measures had been implemented in daily practice, a meeting was organized for the same participants a year after the course.

For the orthoptists in the low-SES areas, we invited hospital managers of clinics to participate in the interactive debates to realize these measures in their practices.

Distribution of the educational cartoon nationwide

We sought ways whereby the educational cartoon could be provided permanently on a nationwide basis. Although several patch manufacturers were interested in buying it, we feared that commercial interests: for example, if a patch manufacturer demanded exclusive rights it, it would no longer be possible to guarantee continued distribution to the children who needed it most: those of parents who spoke little or no Dutch. We therefore aimed to organize its distribution collectively through the department of Technology Transfer at Erasmus University Medical Centre Rotterdam, a semi-governmental institution.

Integration of education on compliance into the training of orthoptists

Finally, we investigated whether education on compliance could be integrated into the primary training of orthoptic students and into the continuing training for working orthoptists. The department of Orthoptics at the University of Applied Sciences in Utrecht ("the

School of Orthoptics") is the only institute in The Netherlands that offers a Bachelor's-level course in Orthoptics; it also provides most continuing-education courses for Dutch orthoptists.

As lecturers who teach the modules on amblyopia at the School of Orthoptics participated in the study committee, it was planned that the modules taught in our course would be incorporated as a course on compliance within the new curriculum for orthoptic students, which was then being reviewed. At the end of the study, we evaluated whether the curriculum for orthoptic students contained modules on noncompliance issues and whether working orthoptists' attendance in continuing education on compliance.

Data collection

Before and after implementation, we observed participants in their practices, and used face-to-face semi-structured interviews and structured questionnaires (Fig. 9.1) assess their conceptions, attitudes and awareness regarding noncompliance, the actions they undertook to deal with noncompliance, and their communication with patients. These instruments were developed on the basis of the principles of Prochaska & Velicer's Stages of Change model (Prochaska et al. 1997) and of the phases in the process of change for care providers and teams described by Grol & Wensing (Grol et al. 2004).

The data collected before implementation was compared with data collected after it. To detect any general changes in orthoptists' attitude to noncompliance, as well as their concepts of it and methods of dealing with, we sent an annual national compliance survey to all Dutch orthoptists over a four-year period, thereby collecting reference data. Detailed descriptions of the instruments used to collect our data and the statistical analysis are described in Appendix XI.

Outcome measures

The outcome measures were utilization of the cartoon, orthoptists' awareness, attitude, and activity in dealing with noncompliance. Secondary, we assessed their patient contact, in particular towards patients whose fluency in Dutch was poor, as compliance with occlusion therapy was especially poor among immigrants who speak Dutch poorly.

RESULTS

Participants

We recruited nine orthoptists working at five hospitals located in non-native, low-SES areas in the cities of Amsterdam, The Hague, Utrecht and in the suburban areas of Rotterdam.

Of all clinics in low-SES areas of the four large cities in The Netherlands approached, only one in Rotterdam refused to participate as according to the orthoptists of these clinic their patients were very compliant (Appendix IV). Elsewhere in NL, 29 orthoptists were willing to participate in the study. Four of these did not receive approval from the supervisory board of their clinics to participate the study. One was excluded as she was a member of the study committee; another because she was a lecturer at the School of Orthoptists where training on compliance was implemented separately. The remaining 23 orthoptists who had been recruited in the study worked in 15 clinics throughout NL (Appendix IV).

Use of the educational cartoon

After implementation, most of the orthoptists distributed the cartoon to all newly diagnosed children with amblyopia who were being treated with occlusion therapy for the first time.

The nine orthoptists in low-SES areas estimated that, per year, they newly treated approximately 143 amblyopic children between 3 and 6 years with occlusion therapy. In a period of 15 months, before implementation, 137 such children were reported to the study centre; in a period of 12 months, after implementation, the orthoptists in these areas distributed the educational cartoon to 79 such children who started occlusion therapy for the first time, and reported these children to the study centre.

Elsewhere in NL, 23 orthoptists estimated that, per year, they newly treated approximately 376 amblyopic children between the age of 3 and 6 years with occlusion therapy. In a period of 15 months, before implementation, they reported 404 such children to the study centre. In a period of 12 months, after implementation, these orthoptists distributed the educational cartoon to 359 such children who started occlusion therapy for the first time, and reported these children to the study centre.

Although the interviews and questionnaires completed before and after implementation did not identify any change in orthoptists' attitudes or methods regarding noncompliance and their approach to it. Orthoptists did evaluate themselves as being more aware of noncompliance, but this change was temporarily. Observations in practice showed that orthoptists spent twice as much time addressing a child after implementation than before it (10″ vs. 24″ in low-SES areas, elsewhere: 26″ vs. 40″). Detailed results from the interviews and questionnaires on the orthoptists' awareness, attitude, and activity in dealing with noncompliance, as well as the results from the observations in practice on the actions taken to communicate with parent and child are described in Appendix XII.

Additional compliance-enhancing measures

Few of the additional compliance-enhancing measures which were planned during the interactive debates in the course on compliance were used in practice; even when they *were* used, it was by few of the orthoptists:

- Six of the 32 orthoptists reported making telephone calls more often to parents of their patients who did not show up for appointments, after implementation.
- A bill was sent to patients who did not show up for appointments after implementation in one clinic.
- The 'Courtesy Call' was introduced at one orthoptist's practice,. This was a letter sent to patients' parents two weeks beforehand to remind of their appointment .
- The time allotted for a patient's first visit did not change after implementation, despite the fact that 24 out of 32 orthoptists had indicated in the structured questionnaire before the implementation that they 'quite often' to 'always' lacked the time to organize extra activities. By observing the participating orthoptists in practice, we found that the time allotted for a patient's first visit was 21′ in low-SES areas and 27′24″ elsewhere in NL (Table 9.1).
- Registering and requesting feedback on children who had been referred for a positive vision screening test at the child healthcare centres were not implemented.
- Neither organizing information sessions for parents of children who need to patch for the first time,
- Nor using more often interpreters with parents who speak Dutch poorly had been carried out since.

Distribution of the educational cartoon nationwide

We chose, for nationwide distribution of the cartoon, the department of Technology Transfer at Erasmus University Medical Centre Rotterdam, a semi-governmental institution experienced in the valorisation of new medical interventions. At cost price, □3.00, the manufacturers were offered the education cartoon, fitted with the company's logo. One company has since made the cartoon available for distribution together with occlusion patches for delivery by pharmacists and orthoptists in The Netherlands, Germany and Italy.

Integration of education on compliance into the training of orthoptists

During the implementation study, new insights and knowledge on compliance (such as that derived from the course on compliance) was hardly integrated into the primary training of orthoptic students, despite the participation in the study of two lecturers from the Department of Orthoptics at the School of Orthoptics in Utrecht.

In retrospect, our strategy for integrating such teaching on compliance into the curriculum of orthoptic students was ill chosen: the School of Orthoptics was under no obligation to

Table 9.1) Results of the observations at the participating orthoptists' practices, subdivided by whether they worked in low-SES areas of the four large cities or elsewhere in NL.

	Low-SES areas (n = 9 orthoptists)			**Elsewhere in NL** (n = 23 orthoptists)		
	Pre-implementation	**Post-implementation**	*P*-value	**Pre-implementation**	**Post-implementation**	*P*-value
Number of patients observed	132	73		344	198	
Appointments missed without cancellation	14.7%	12.0%	0.600	5.3%	8.5%	
Mean allotted time (minutes) per new visit †	21:00 SD 9:57	20:00 SD 4:37	0.741	27:24 SD 6:44	26:57 SD 6:10	0.328
Mean allotted time (minutes) per follow-up visit †	15:37 SD 1:46	14:22 SD 3:12	0.170	16:44 SD 2:26	16:31 SD 2:21	0.327
Parental fluency in Dutch †						
Excellent	50.1%	34.8%	0.133	88.3%	87.4%	0.811
Good	28.9%	30.0%	0.913	5.1%	9.4%	0.142
Moderate	19.9%	32.4%	0.077	3.9%	2.2%	0.426
Poor to none	3.1%	3.9%	0.861	2.4%	0.9%	0.143
Length of orthoptist's explanation (minutes) †‡						
to the parent	2:30 SD 1:04	2:45 SD 2:12	0.832	2:51 SD 2:04	3:06 SD 2:09	0.796
to the child	0:10 SD 0:11	0:24 SD 0:33	0.061	0:26 SD 0:43	0:40 SD 1:13	0.121
How the explanation was given *†						
through compulsory formulations §	73.9%	46.9%	**0.050**	73.7%	43.7%	**<0.001**
through appreciative formulations ⊡	73.9%	66.1%	0.528	74.4%	70.4%	0.654
Repeated the explanation twice or more †	34.1%	57.3%	**0.036**	30.8%	22.3%	0.073
Tools used to clarify explanation†						
Pictures, posters or statuettes	2.4%	0%	0.080	1.9%	0.8%	0.392
Drew figures during explanation	1.6%	1.8%	0.997	1.7%	7.4%	**0.022**
Patient's family member or friend translated	0.9%	0%	0.351	1.4%	0.4%	0.254
Official medical interpreter translated	0.0%	1.8%	0.352	0.0%	0.0%	-
Nothing	97.4%	96.4%	0.948	95.0%	91.3%	0.210
Verified whether explanation was understood †						
Yes: "Yes?" or "Okay?"	26.1%	40.1%	0.176	44.4%	34.7%	**0.007**
Yes: "Do you understand?" #	4.6%	19.3%	**0.068**	3.0%	21.5%	**<0.001**
No	69.3%	40.6%	**0.012**	52.6%	43.8%	0.399

* P < 0.05

† Patients ≤ 12 years who were observed by the researcher

‡ Only explanations on occlusion therapy, § For example: "You must...", "You need...", "Remember that you…", © For example: "the best is...", "it is important...", "I can imagine...", "try to…", "I advise you to…", # Other formulations used: "Do you have any questions?", "Is there something else that you want to know?"

integrate compliance issues into the curriculum for the basic training for orthoptists. The need for such changes is usually expressed by a governmental body. In such an event, the orthoptists' professional organization would formulate targets for professional training that would be implemented by the school.

The course on compliance was provided by a lecturer from the School of Orthoptics, who normally arranged sessions on research methodology. In principle, it was to be provided three times as a continuing course to all orthoptists in The Netherlands. However, due to insufficient participant numbers (fewer than 20 registrations), it was cancelled on all three occasions. No participants registered for the first course; four for the second, and seven for the third.

DISCUSSION

We found that the educational cartoon that explains without words to a four-year old child why it should wear the eye patch was used best in low-SES areas where many patients speak Dutch poorly. It was adopted by all orthoptists in low-SES areas, against only two-thirds of those in other areas. Awareness of noncompliance among orthoptists changed temporarily. Hardly any of the additional compliance-enhancing measures that were planned, were implemented, although the study group and participating orthoptists had conceived and planned these with the best intentions. The integration of training on compliance into the curriculum for student orthoptists failed, as did the continuing course on compliance for working orthoptists. On the other hand, the nationwide distribution of the cartoon is well organized by the department of Technology Transfer at Erasmus University Medical Centre Rotterdam, and supported by companies that manufacture eye patches.

In another part of the study (see Chapter 7 of this thesis), we reported on the effectiveness of the cartoon in terms of electronically measured compliance with occlusion therapy of children living in the low-SES areas. Whereas mean electronically measured compliance was 52% before implementation, their treating orthoptists estimated it to be 74%. After implementation, compliance was 62%, whereas their treating orthoptists estimated it to be 55%.

In contrast with these low figures, our observations of the orthoptists in their practices in the current study showed that only a quarter of them suspected noncompliance only in a single patient during one day of observation; three quarters never suspected noncompliance. How can orthoptists' low awareness and estimates of noncompliance be explained?

As occlusion treatment is very effective, and visual acuity may still rise even if compliance is as low as, for example, 30%, it is possible that an orthoptist does not notice cases of moderate compliance. Children with moderate compliance will eventually reach sufficient visual acuity (Stewart et al. 2004b); the only difference is that it will take them longer to do so. Stewart and colleagues (Stewart et al. 2004b) found in their study that dose rates of 2 to 6 hours/day, measured objectively with the ODM, generate equal final outcomes, although those with a high dose rate achieved a successful outcome more rapidly.

Changes in awareness were most pronounced in the period before and after the training course: according to the questionnaires, the orthoptists reported being more aware of noncompliance, and estimated that patients' compliance was lower than before implementation. It seems that the attention raised by the implementation phase and the course may have caused this temporary change. Various other implementation studies have reported similar findings with regard to the implementation of products, guidelines or protocols in healthcare practices (Grol et al. 2003; Grol et al. 2005). In literature, it was suggested that passive dissemination (e.g., mailing educational materials to targeted clinicians) is unlikely to result in a maintained behaviour change when used alone. A multifaceted intervention with education, guidance and evaluations before, during and after implementation until the long-lasting implementation of the product in practice in was best effective (Cabana et al. 1999; Grimshaw et al. 2004; Grimshaw et al. 2002; Grol et al. 2004; Grol et al. 2005; Horner et al. 2004).

Good compliance depends on good communication: Orthoptists verified more often whether their explanations had been understood, and also repeated these explanations more often, especially in low-SES areas, after implementation. The time they spent in explanations to the child had also changed: they addressed the child twice as much as before. However, this was still less than a minute – only 15% of the time spent in explanations to the parent.

This study has its limitations. First, the presence of the researcher may have influenced the orthoptists' everyday behaviour during the observations at their practices. In spite of this, only a quarter of orthoptists suspected noncompliance each in just one patient during one day of observation. Secondly, the questionnaires were not validated, as this was not possible in this small scale study and no questionnaires exist to measure these specific outcomes. Thirdly, the 23 orthoptists elsewhere in NL volunteered to take part in the study: they may have been eager to adopt the cartoon in their practice. In spite of this, not all of them used the cartoon.

It is remarkable that hardly any additional measures to enhance compliance were implemented in the orthoptic practices, even though these measures had been conceived and planned by the study group and by the participating orthoptists with a lot of conviction and the best intentions. These measures demanded extra, unpaid, time, effort or investment by the orthoptist or by the hospital – resources that are especially scarce in hospitals in low-SES areas where the cartoon is most needed.

Acknowledgments

The authors thank the following lecturers for their voluntary contribution to the course on compliance: Dr. S. Simons, Department of Clinical Pharmacy Institute of Pharmacy, University of Bonn; Dr. E.R. Heerdink, Department of Pharmacoepidemiology & Pharmacotherapy, Utrecht University; Dr. J.H. Groenewoud, Expertise Centre for Transitions of Care, Rotterdam University of Applied Sciences; Mrs. L. Peereboom, lecturer in communication skills, Utrecht University of Applied Sciences; and Mrs. H. van Agt, Department of Public Health , Erasmus University Medical Centre Rotterdam (Erasmus MC).

Our thanks are also due to the staff at the Department of Technology Transfer at Erasmus MC in Rotterdam for their work on valorising the educational cartoon nationwide.

The authors also wish to thank the 20 clinics for their contribution to the study, and particularly the 32 participating orthoptists, who are listed here first in alphabetical order of clinic and surname:

Amphia Hospital, Breda & Oosterhout (K. Croft, C. Klerks-Kuijpers, P. Neomagus, C. Stoop); Catharina Hospital, Eindhoven (E. van Rooijen-Troost); Flevohospital, Almere (L. Bakker, N. Bos-De Bruin); Isala Clinics, Zwolle (A. Wellner-Fokker); Kennemer Hospice, Haarlem (W. Jutte, A. Hupsch-Van den Heuvel, A. Spaan); Lievensberg Hospital, Bergen op Zoom (M. de Rooij, A. van Voorden-Van der Knaap); Máxima Medical Centre, Eindhoven & Veldhoven (E. Cockx-Huitema); Medical Clinic, Alkmaar (W. Hoogeveen, I. Huisman); Medical Centre Haaglanden, The Hague (E. van Minderhout, B. Simonsz-Tóth, M. Vermeulen-Jongen); Mesos Medical Centre, Utrecht (A. van de Bovenkamp-Scheper, A. Versteeg); Rivierenland Hospital, Tiel (T. Kooij-'t Spijker); Ruwaard van Puttenhospital, Spijkenisse (M. Kruiswijk); Sint Lucas Andreas Hospital, Amsterdam (A. Burger-Meywaard); Tergooi Hospitals, Hilversum (D. Hamers); Twenteborghospital, Almelo (I. 't Hart-Leemhuis, M. Hazenkamp-Boekhold, F. Jansen, D. van der Nagel-Pronk); Vlietland Hospital, Vlaardingen (S. Rousse, D. Valster); Zweeds Rode Kruis Hospital, Zierikzee, Van Weel-Bethesda Ziekenhuis, Dirksland, & Medical Centre, Middelburg (J. Kieviet-De Geus). We also thank all Dutch orthoptists for filling in the questionnaire.

Special thanks for his advice go to Prof. G. Van Rij of the Department of Ophthalmology at Erasmus MC Rotterdam. Acknowledgements for proofreading and correction of the English text are due to Mr. D.R.M. Alexander of Ways with Words, Mrs. K. Verhoef of VERT-ICAL Translations & Administration and Mrs. A. Van Rij-De Jong.

Chapter 10

General Discussion

GENERAL DISCUSSION

Noncompliance limits the effectiveness of detection and treatment for amblyopia. As described in Chapter 1, amblyopia is the most common visual defect in children, which can be treated effectively when treatment is started at a young age. However, one third of the children affected do not reach a visual acuity of 0.5 (i.e. 0.3 logMAR) in their amblyopic eye, mainly because they do not comply with the occlusion hours prescribed by the orthoptist. By increasing the risk of bilateral visual impairment through loss of vision in the non-amblyopic eye, this noncompliance reduces their quality of life in adulthood.

Early detection of Amblyopia

The contribution of the Dutch child vision screening program up to age 7 to the detection of amblyopia was assessed in the Rotterdam Amblyopia Screening Effectiveness Study (RAMSES) (Juttmann 2001). We found that due mainly to the visual acuity measurements performed after the age of 3, the screening programme detected half of the children with amblyopia (Chapter 2). Preverbal screening – which included the corneal light reflex, cover-uncover test, observation of ocular pursuit movements, inspection of the cornea and pupil, and a pupillary light reflex test – contributed little to the detection of refractive amblyopia.

The prevalence of amblyopia was 3.4%, which is consistent with previous estimates, which ranged from 0.02% to 5.3% (Attebo et al. 1998; Friedman et al. 2009b; Multi-ethnic Pediatric Eye Disease Study Group 2008; Robaei et al. 2008; Robaei et al. 2006; Webber et al. 2005). At age 7, a quarter of amblyopic children (0.8% of the total population) had visual acuity in the worse eye of <0.5 (i.e. >0.3 logMAR). In a sample of 6-year-old Australian children this percentage was 0.7% (Robaei et al. 2006); in a screened population of 12- to 13-year-old children in Sweden it was 1.1% (Ohlsson et al. 2001).

In the RAMSES, the mean presenting age to the orthoptist or ophthalmologist was 2.3 years for strabismic amblyopia, 2.5 years for combined-mechanism amblyopia, and 4.5 years for refractive amblyopia. In the late 1960s, before the national implementation of vision screening in The Netherlands, the various types of amblyopia were detected more than 2 years later: 5.1 years for strabismic amblyopia, 5.7 years for combined-mechanism amblyopia, and 6.6 years for anisometropic amblyopia (Simonsz-Toth et al. 2007).

On the basis of the results from the RAMSES, we suggest that the greatest contribution to amblyopia detection is made by preschool screening from age 3. Preschool visual acuity measurements from age 3 played, in particular, an important role in the detection of amblyopia, especially of refractive amblyopia. Cases of strabismic or combined-mechanism

amblyopia were relatively more likely to be self-referred than were those of refractive amblyopia. It remains to be seen whether earlier detection and treatment of amblyopia is more effective (and cost-effective).

The RAMSES data also show that one-fourth of the positively screened children in the vision screening programme were referred unsuccessfully (Chapter 3). This kind of non-compliance involved screening physicians and parents alike, the four main reasons for unsuccessful referral with vision screening for amblyopia being parents' failure to comply with referral, miscommunication that led to failed referrals after an abnormal screening test, and doctors' poor documentation and failure to follow the referral guidelines. The impact of unsuccessful referral in terms of visual acuity outcome was small, although not negligible: 3 out of the 16 cases who had insufficiently treated amblyopia in the birth cohort were due to unsuccessful referral.

In the RAMSES study, differences in socioeconomic status between the children who had been referred successfully and those who had been referred unsuccessfully nearly reached statistical significance. Other studies are more conclusive in this regard (Loudon et al. 2006; Mark et al. 1999; Subramanian et al. 2004; Sutton et al. 1994; Vernon et al. 1990; Williamson et al. 1995). In one Dutch study, it was also apparent that parents of non-Western children were less likely to consult an ophthalmologist than parents of Dutch children (Van Laar 2007).

In some of the cases in which unsuccessful referral was due to parents' unawareness of the referral, no written evidence of any referral could be found in the children´s screening records at the child healthcare centre, even though a positive screening test had been reported to the study centre. We suggest that the screening physicians in such cases may have deviated from the screening protocol, especially when visual acuity at screening was near the referral threshold. Many of the children who had been referred unsuccessfully had slightly low visual acuity at screening. One can imagine that a physician or parent's decision not to act upon a positive vision-screening test might have been influenced by a normal (negative) screening history, normal subsequent screening examinations, or the absence of clinical symptoms of visual impairment.

Compliance in Amblyopia treatment

Although various studies have examined how noncompliance with occlusion therapy for amblyopia manifests itself in children or their parents (Awan et al. 2005; Awan et al. 2010; Dixon-Woods et al. 2006; Dorey et al. 2001; El-Ghrably et al. 2007; Karlica et al. 2009; Loudon et al. 2006; Loudon et al. 2009; Loudon et al. 2003; Loudon et al. 2007b; Stewart et al. 2005; Stewart et al. 2004b), they have not explored how orthoptists deal with it. Be-

cause it has been shown that it is difficult for healthcare professionals to identify patients with poor compliance (Copher et al. 2010; Heszen-Klemens 1987; Kass et al. 1986a; Russell et al. 2003; Urquhart 1997), we listed Dutch orthoptists' conception, awareness and attitudes of noncompliance, and the action they undertook to deal with it (Chapter 4).

The considerable variety we found is presented in Chapter 4. Orthoptists were aware that compliance is an important issue in their current practices, and were eager to learn more about structured methods for detecting and dealing with it. This finding contrasted with descriptions in earlier studies evaluating healthcare professionals' management of noncompliant behaviour, which showed that noncompliance was sometimes ignored completely (Gelb et al. 2008; Heszen-Klemens 1987; Wright 1993), largely because it conflicted with professionals' beliefs, norms and expectations regarding the roles they deemed to be "proper" to patients and professionals (Hood 2008; Playle et al. 1998; Stimson 1974; Wens et al. 2005).

Although orthoptists stated that compliance was an important issue within orthoptic practice, they estimated their own patients' compliance to be 73.8% – rather high relative to the mean electronically measured compliance with occlusion therapy found in other studies (48%-57% (Awan et al. 2005; Loudon et al. 2006; Stewart et al. 2004b)). During our observations at the practices of the participant orthoptists, we also noted that it seldom occurred to an orthoptist that his or her patient had not patched. During a day's observation, only a quarter of them suspected noncompliance, and then only in a single patient. The others suspected nothing.

Other studies have reported on this disparity between actual and assumed levels of compliance (Copher et al. 2010; Kass et al. 1986a; Moreau et al. 2009; Wylie et al. 2002), most healthcare professionals identifying defaulting patients on the basis of the results of therapy. Much of this prediction of a patient's compliance was based on their own instinctive feeling (Gelb et al. 2008; Kass et al. 1986a; Wens et al. 2005). It is therefore likely that orthoptists start to worry about their patients' compliance only when visual acuity in the amblyopic eye does not increase – for it has been shown that occlusion therapy is very effective: increases in visual acuity will cease only if compliance is very low. Stewart et al describe a randomised control trial in which children were prescribed either 6 or 12 occlusion hours per day (Stewart et al. 2007b). Even though children who patched more reached good visual acuity more rapidly, visual acuity improved in all those who patched for more than 2 hours a day. As children with moderate compliance take longer to reach sufficient visual acuity, it is therefore hard to detect moderate compliant behaviour in those who patch for amblyopia.

In general terms, occlusion therapy for amblyopia is a long and exhausting therapy – for children and parents alike. It is probably unavoidable that, accidently or deliberately, therapy is missed occasionally (Nevins 2005). Compliance with any treatment or medication is a complex phenomenon (Lehane et al. 2009), and is affected by a range of factors, especially communication, which is itself essential to compliance (Conn et al. 2007; Friedman et al. 2008). Earlier research in The Hague (The Netherlands) by Loudon and colleagues has shown that compliance with occlusion therapy was especially poor in children of parents who spoke poorly Dutch, who were poorly educated, and/or came from another country (Loudon et al. 2006). The most important clinical parameter was the initial visual acuity of the amblyopic eye poor visual acuity. It was suggested that the acceptance of the patch on the good eye is less when acuity is poor in the amblyopic eye (Awan et al. 2010; Hiscox et al. 1992; Lithander et al. 1991; Loudon et al. 2006; Loudon et al. 2003; Smith et al. 1995; Stewart et al. 2005; Stewart et al. 2007b).

Factors affecting compliance

In addition to what is known from the literature, we found that poor compliance with occlusion therapy in a group of children from ethnically diverse, low-SES areas seemed to be correlated with indicators of social cohesion (Chapter 6). Low compliance was associated not only with a high degree of social cohesion at the micro level (family, neighbours and friends), but also with low social cohesion at the macro level (Dutch society). Compliance was also low when parents had depression or a low income.

It was difficult for us to interpret these new findings. Although the parents' fluency in Dutch did not correlate with compliance in this study sample, it may have influenced the way these factors were correlated with compliance. An inability to speak Dutch necessitates family bonds and neighbour contacts (Schouten et al. 2006). As poor fluency in Dutch is likely to reduce the chance of finding work, any poverty that results is equally likely to induce depression (Gonzalez-Castro et al. 2011), and thereby to affect social cohesion (Fassaert et al. 2011). Parent's inability to speak Dutch may also cause the orthoptist's instructions to be misunderstood (Harmsen et al. 2003; Harmsen et al. 2006; Loudon et al. 2006; Van Wieringen et al. 2002). It is therefore hard to conclude exactly which of these factors caused noncompliance.

In the same group of children from ethnically diverse, low-SES areas, low patching was also associated with noncompliance when patching interfered with the child's outdoor activities (Chapter 6). The literature has shown that a degree of distress and stigma are related to occlusion therapy, and that past compliance behaviour was predictive for future compliance behaviour (Dixon-Woods et al. 2006; Felius et al. 2010; Holmes et al. 2003;

Loudon et al. 2009; Parkes 2001). In this study sample, our questionnaire could not find similar psychosocial factors that involved compliance with the therapy.

In the last few years, social and political affairs in The Netherlands have been somewhat overshadowed by assertions about the perceived failures of the multicultural society and the role that religion supposedly plays in them. Against this background, it is therefore interesting to report that we found no evidence that religion had any effect on compliance with occlusion therapy. Overall, the literature does not show whether religion has any clear bearing on healthcare behaviour; there may thus be either a positive or negative influence (Hjelm et al. 2003). For example, a study among HIV patients showed that certain religious practices were positively associated with treatment adherence, whereas, due to the stigma attached to HIV, other religious beliefs played a negative role (Parsons et al. 2006).

Neither, with regard to less emotive issues, we investigated the relationship between compliance and different brands of eye patch. In Chapter 5, we described the differences between the comfort and mechanical properties of the four most common brands: 3M (Opticlude), Master-Aid (Ortopad), Lohmann-Rauscher (Pro-ophtha) and BSN Medical (coverlet S), and the influences of these factors on compliance. This produced large differences between the brands in terms of skin reaction, discomfort when removing the patch, and the cosmetic and adhesive strengths of the patches. At room temperature, the breathing characteristics of all patches were minimal; in the sun, some could be compared to wearing a raincoat, and would therefore be better suited for shorter periods of occlusion.

There were also considerable differences in the strength needed to remove a patch from the skin. As the maximum force ranged between 2.6 (Lohmann-Rauscher) and 8.8 (BSN Medical) Newton, certain brands of patches would clearly be more suitable for a longer duration of patching. We therefore suggest that the most important factor influencing wearing comfort is the glue layer, which, by determining the degree of force needed to remove the patch, can have a bearing on any irritation of the skin.

Significant associations between compliance and different brands of eye patch could not be found, however. Nevertheless, we suggest that it seems reasonable to expect orthoptists to consider comfort of wear when prescribing a certain brand of patch, and for manufacturers to spend more time and effort on improving the properties of their patches, especially those affecting the glue layer.

Improving compliance

In a randomised control trial in The Netherlands, electronically measured compliance with occlusion therapy was greatly improved by an educational cartoon made by José Vingerling and Gerard de Bruyne. But even when a product, guideline or protocol proves effective, its implementation in daily practice does not follow automatically.

The last part of this thesis describes the results of the implementation of the educational cartoon (see Chapters 7 and 9). As stated above, this cartoon can increase compliance with occlusion therapy for amblyopia. Over recent decades, however, more has been learned about the difficulties of getting evidence-based products into "real" daily practice, where they are often different or entirely absent (Cabana et al. 1999; Glasgow et al. 2003; Grimshaw et al. 2004; Grimshaw et al. 2002; Grol et al. 1999; Grol et al. 2005; Horner et al. 2004; Stetler et al. 2008).

Chapter 7 states that positive effects were found after implementation of the educational cartoon in daily orthoptic practice: electronically measured compliance in children living in low-SES areas was slightly higher in the group that received the cartoon than in the group that did not (62% vs. 52%). Although this finding was less pronounced than that in the previous randomised control trail, which was performed in The Hague by Loudon and colleagues (Loudon et al. 2006), the proportion of poorly compliant children in our study (which was performed in low-SES areas) was greater: in low-SES areas, 42% of children who did not use the cartoon patched less than 30% of the occlusion time prescribed, against 15% in The Hague (Loudon et al. 2006).

Nonetheless, despite this small effect on compliance, we were able to demonstrate an improvement in the clinical relevant outcome measure: after implementation, the difference in acuity between both eyes decreased more rapidly during occlusion treatment than before implementation (3 lines of acuity per year vs. 2.4 lines per year). This means that treatment lasted less long when the cartoon was used. These advantages in SRIAD remained after adjustment for its confounding factors (mother's fluency in Dutch, age at start of occlusion therapy, initial intraocular visual-acuity difference, and cause of amblyopia). The SRIAD was higher in severe amblyopia than in mild amblyopia, partly because orthoptists patched more in severely amblyopic children. The SRIAD was lower in older children, despite the fact that orthoptists patched more in these children. The relationship between age and intraocular visual acuity at start of occlusion therapy, and SRIAD for the entire study population was as follows: *SRIAD = – 0.01938 – 0.5669 x initial intraocular visual-acuity difference + 0.00427 x age*.

The speed of the reduction in intraocular acuity difference was higher after implementation, especially among children living in low-SES areas, in whom, on average, the difference between acuity of the amblyopic eye and acuity in the healthy eye decreased by 2.2 lines per year before implementation, against 3.2 lines per year after implementation. In other areas of The Netherlands, it decreased by 2.4 before implementation and 2.9 afterwards. Similarly, attendance rate improved considerably in low-SES areas and not at all in areas elsewhere in NL. However, these effects may have been due partly to additional measures (such as the course on compliance) taken at the same time as the educational cartoon.

Chapter 9 describes how the implementation of the cartoon affected the orthoptists' awareness of compliance behaviour, as well as their attitude towards it and management of it. We showed that the cartoon is probably best used in low-SES areas where many of the patients speak Dutch poorly. All orthoptists in low-SES areas adopted it, against only two-thirds of their colleagues in other areas – which is consistent with the finding that the improvement in SRIAD was stronger in low-SES areas than elsewhere in the NL.

As described in Chapter 4, orthoptists found compliance to be an important issue in their daily practice, and were eager to learn more about it and about how to improve it. It is therefore interesting that orthoptists in low-SES areas overestimated their patients' compliance before implementation. After implementation, they estimated it to be lower than objectively recorded electronically: at 54%, against an electronically measured compliance of 62%. This indicates precisely what was described earlier: it is difficult to estimate patients' compliance.

After implementation, orthoptists reported being more aware of noncompliance, these changes in awareness being pronounced in the periods before and after the training course. This seems to have been the product of the attention raised by the implementation phase and the course. Overall, the orthoptists' ability to suspect specific cases of noncompliance was not greater after implementation: in an entire day of observation, only a quarter of them suspected it, and then only in a single patient.

Something else that improved after implementation was communication: orthoptists verified more often whether their explanation had been understood, and repeated their explanations more, especially in low-SES areas. After implementation, they devoted twice as much time to the child as they had before. Nevertheless, this still totalled less than a minute – only 15% of the time they spent explaining to the parent. Neither, after implementation, did they allot more time to consultations with patients who did not speak Dutch. It is ironic that the time allotted for a first visit in low-SES areas was 20% less than

that for a first visit elsewhere in The Netherlands, especially as most of that time would be needed to ensure good communication between orthoptist, parent and child.

In The Netherlands, each person giving healthcare is legally responsible for clearly explaining the diagnosis and therapy to their patients, irrespective of these patients' situation. To children under the age of 12, the explanation should be given in such way that best suits the child's comprehension (Department of Justice 1994). At this point we should note that time may be becoming an increasingly scarce commodity for a practicing orthoptist: as Winnick and colleagues observed (Winnick et al. 2005), paediatric practices now have to manage full bookings and carry out their usual administrative and billing activities in less time. This may also be the case in orthoptic practice: market forces – which are becoming more and more apparent in the Dutch healthcare system – may well be reducing the amount of time an orthoptist can devote to addressing the needs of patients and their families during clinical visits. We noted in our study that although the study group and participating orthoptists had the best intentions when conceiving and planning the additional compliance-improving measures, hardly any of these measures were eventually achieved.

Similarly, attempts to integrate education on compliance into the curriculum for student orthoptists and into the continuing course on compliance for working orthoptists were unsuccessful. These measures seem to require extra time, effort or investments on the part of orthoptists and hospitals – resources which seem to be scarce in practices nowadays, as we have just stated above. Without managerial incentives or sanction, it is difficult to implement permanent compliance-improvement measures in daily orthoptic practice, as there appears to be a conflict of interest between public health and running a hospital or practice.

Nevertheless, as the cartoon is clinically a relevant product that achieves good treatment outcomes, it is being made available through the department of Technology Transfer at Erasmus MC Rotterdam, The Netherlands' largest teaching hospital, which is experienced in the valorisation of novel medical interventions. The department organises the nationwide distribution of the cartoon, which is supported by manufacturers of eye patches. The cartoon is now available along with occlusion patches for delivery by pharmacists and orthoptists, who distribute patches directly to the child.

Finally, we found that the advantages of the cartoon were especially pronounced in children in low-SES areas with a large proportion of immigrants who spoke Dutch poorly. We suggest that this was due to the direct explanation of occlusion therapy to the child. We therefore performed a new randomised study that involved the three elements of the

educational cartoon in low-SES areas (Chapter 8). This demonstrated that the greatest improvement in compliance with occlusion therapy was brought by the educational cartoon, not by the calendar with reward stickers or by the parent-information leaflet. Several other studies have shown that more direct communication between physician and child helps improve satisfaction with care, compliance with treatment, and health outcomes (Holzheimer et al. 1998; Pantell et al. 1982; Tates et al. 2001). To this it should be added that children understand more about concepts of health and illness than is generally assumed (Holzheimer et al. 1998; Lewis et al. 1984; Tates et al. 2001).

Measuring treatment outcome by measuring the speed of success

Another innovation in this thesis is the fact that we measured the effectiveness of the cartoon's implementation on the basis of the rate of decrease in visual-acuity difference between the amblyopic eye and fellow eye ("Speed of Reduction in Intraocular-Acuity Difference"; SRIAD).

Defining and measuring unilateral amblyopia treatment outcome in a research setting has been described as a major challenge, and various methods have been used to achieve it. The literature has provided several definitions of treatment success, including 1) the achievement of a specific acuity by the end of the treatment period (e.g. 6/6 (i.e. 0.0 logMAR) or 6/9 (i.e. 0.2 logMAR); 2) equal visual acuity in the amblyopic and fellow eyes; and 3) proportional improvement (i.e. log unit change in the visual acuity of the amblyopic eye divided by the difference between the amblyopic eye at the start of treatment and the fellow eye at the end of treatment) (Epelbaum et al. 1993; Flynn et al. 1978; Fulton et al. 1988; Hiscox et al. 1992; Mintz-Hittner et al. 2000; Repka et al. 2008; Stewart et al. 2003; The Paediatric Eye Disease Investigator Group 2002; Thompson et al. 1996). Among the disadvantages of these approaches is that they did not account for the severity of amblyopia at the start of treatment, account for the amount of improvement during the treatment, or consider the acuity in the fellow eye at both the start and the end of the treatment (Stewart et al. 2003).

To overcome these limitations, we used the SRIAD as a measure of the effectiveness of the educational cartoon. This approach also enabled us to calculate the treatment outcome while adjusting for confounders influencing the visual-acuity outcome. It is therefore suggested that this approach (i.e. calculating the outcome in the speed of treatment success) might therefore be useful to quantifying treatment outcome in future studies.

CONCLUSIONS

According to the RAMSES data, the Dutch vision screening programme – which consists of eight consecutive screening examinations between the ages of 0 and 6 – contributes to at least two year earlier detection of amblyopia than when compared with the age of detection in a Dutch historical cohort which did not receive vision screening in childhood. Preschool screening from age 3 contributes the most to amblyopia detection.

Effective detection of amblyopia depends not only on an effective screening program, but also on good compliance with referral after a positive screening test. Although, the impact of unsuccessful referral in terms of visual acuity outcome was small, it remains interesting that screening physicians as well as parents were involved in this kind of noncompliance.

While there is a need to determine the effectiveness of a reduced-vision screening programme and the cost-effectiveness of vision screening in general, the overall efficacy of screening might be improved by an effective monitoring feedback system after referral.

Also the effectiveness of amblyopia treatment is influenced by poor compliance. However, in our study, compliance in daily practice is hard to notice for orthoptists, despite the fact that these orthoptists were aware that compliance is an important issue in their current practices.

In case of noncompliance with occlusion therapy, orthoptists have a variety of practice-based actions and methods to deal with this noncompliance. In addition to this, we found with regard to prescribing occlusion therapy that there were large differences in the comfort and mechanical properties of eye-patches; therefore, when prescribing a certain brand of patch, account should be taken of this variety.

In addition to what is found in literature, good compliance with occlusion therapy is not only influenced by fluency in the native language, country of origin or level of education, but it is among migrants also correlated with a high social cohesion in the Dutch society. In other words, migrants who were integrated in the Dutch society had good compliance.

In the implementation study, the educational cartoon was proven again to be effective in improving compliance with occlusion therapy. It also improved the outpatient attendance rate, visual acuity outcome and duration of treatment. We suggest that this is because the cartoon story explained the purpose of the treatment without any words. In addition, it targeted children directly, helping them to increase their understanding, and simultaneously stimulating them to complete the therapy. As a similar approach could also be taken

to other kinds of therapy in children, we are currently developing a similar cartoon that explains to children why and how they should manage asthma.

With regard to the implementation of compliance-enhancing measures in 'real' daily practice, these measures were hardly implemented although these had been conceived and planned by the participants of the study with a lot of conviction and the best intentions. These measures demanded extra, unpaid, time, effort or investment by the orthoptist or by the hospital – resources that are especially scarce in hospitals in low-SES areas where the cartoon is most needed.

Currently, the nationwide distribution of the cartoon is well organized by the department of Technology Transfer at Erasmus University Medical Centre Rotterdam (a semi-governmental institution experienced in valorising medical interventions), and supported by companies that manufacture eye patches.

Finally, in future amblyopia studies, the rate of decrease in visual-acuity difference in log unit between the amblyopic eye and the fellow eye ("Speed or Reduction in Intraocular-Acuity Difference", SRIAD) might be useful to quantifying treatment outcome.

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Summary & Samenvatting

SUMMARY

With a prevalence of 3-4%, amblyopia (a 'lazy eye') is the commonest eye disorder in childhood. It is a reduction in visual acuity, usually in only one eye, caused by disruption of normal visual development. The acuity in the amblyopic eye can be restored by patching the good eye with an eye patch before the age of six. The success of this treatment depends on several factors, including early detection, start of treatment, and the patient's compliance – i.e., the degree to which the patient follows or completes the therapy prescribed by the caregiver.

As objectified with the Occlusion Dose Monitor (ODM), electronically measured compliance with occlusion therapy is moderate: on average, occlusion is carried out for only 57% of the hours prescribed. In The Netherlands, a randomised control trial showed that compliance was greatly improved by an educational cartoon ("*Het Plakboek*") designed by José Vingerling and Gerard de Bruyne. In a story without words, the cartoon explained to a four-year old child why it should wear the eye patch. The cartoon package also included reward stickers for the children to use on a calendar. For the parents, it included a six-language information sheet about the disease.

Given this knowledge, the aim of this thesis (described in **chapter 1**) was to determine the contribution of Dutch vision screening on the timing of detection of amblyopia, to explore complex compliance issues which may influence the effectiveness of amblyopia treatment, and to evaluate the implementation of the effective educational cartoon in daily practice – for even though a product has been shown to be effective, it is it is not always easy to implement it on a daily basis.

The contribution of Dutch vision screening on the timing of detection of amblyopia was evaluated in the Rotterdam Amblyopia Screening Effectiveness Study (RAMSES). **Chapter 2** describes the clinical results of this follow-up study: strabismic and combined amblyopia were aged about 2.5 years when they first visited the orthoptist or ophthalmologist, thus two years younger than children with refractive amblyopia (4.5 years). In comparison with a cohort that did not received vision screening the various types of amblyopia were detected more than two years later. On the basis of the results from the RAMSES, we suggest that the greatest contribution to amblyopia detection is made by preschool screening from age three. However, due to earlier self-referral to a hospital, or due to unsuccessful referral to a hospital, a quarter of these children did not directly benefit from a positive screening result. We found that unsuccessful referrals after positive vision screening had been caused by parental noncompliance, miscommunication between screening physi-

cian and parent, deficient documentation, and the screening physician's noncompliance with screening guidelines (**chapter 3**).

As poor compliance is a major limiting factor to the effective provision of care, it is interesting to know how healthcare givers (orthoptists) handle noncompliance. In **chapter 4**, we discussed the orthoptists' various attitude and approaches with regard to compliance. This produced three main findings. First, orthoptists underestimated noncompliance with occlusion therapy. Second, they use various practice-based solutions to deal with it. Third, even though noncompliance is particularly high among children of parents who speak Dutch poorly, they devoted less time to explanation in low-socioeconomic status (SES), non-native, deprived areas than they did elsewhere in The Netherlands, where linguistic problems were less common. The time allotted for a patient's new visit was also approximately 7.5 minutes shorter in deprived areas than in other areas.

In addition, several determinants of noncompliance with occlusion therapy were not identified yet. In **chapter 5**, we investigated how compliance might be influenced by the material properties of the four frequently used brands of eye patch and the comfort of wearing these patches. The comfort of each brand of eye patch was moderate. Although there was no difference in electronically measured compliance between patches, there were great differences between the patches with regard to their removal, and also to their cosmetic appearance, opacity and strength of adhesion to the skin. These results suggest that the choice of a brand of eye patch should be based on the number of hours of occlusion prescribed per day.

A previous study has demonstrated that compliance with occlusion therapy was especially low among children of non-native parents who spoke Dutch poorly or had a lower level of education. In **chapter 6,** we determined which immigrant-related factors influenced the degree of compliance. We found that poor compliance was associated with a high degree of social cohesion at micro level (family, neighbours and friends) and a low degree of social cohesion at macro level (Dutch society).

The last part of this thesis focuses on the implementation of the educational cartoon and other compliance-improving measures in daily practice, and the effects these on amblyopia treatment. **Chapter 7** reports on the clinical results of this implementation. Electronically measured compliance and attendance rates were higher in children who lived in deprived areas and had been recruited after implementation than they were in those who had been recruited before implementation. Mean compliance was 52% before implementation and 62.3% afterwards; attendance rates were 60.3% before implementation, and 76% afterwards. After implementation, intraocular acuity difference (SRIAD) improved more rapidly

in children from deprived areas, and also in those from other areas in The Netherlands. In the entire study population, the difference in visual acuity between both eyes decreased by 2.4 acuity lines per year in the group recruited before the implementation, and by 3.0 acuity lines per year in the group recruited after implementation. As these findings were more pronounced in the deprived areas, they support the use of the cartoon in children living in such areas.

With regard to the effect of the educational cartoon itself, this cartoon consisted of three elements (i.e. the cartoon story, calendar with reward stickers, and parental information sheets in six languages). Therefore, it remained unclear which of these increased compliance the most. In a new prospective randomised study (**chapter 8**), the best electronically measured compliance was in the group who received the cartoon story (89%, on average). The second best was in the group who received the information sheets (73%, on average), and the third was in the group receiving the calendar with reward stickers (67%, on average). The lowest compliance was measured in the control group, in which children received a picture to colour (55%, on average).

In **chapter 9**, we demonstrated that the educational cartoon can indeed be implemented in daily practice. It seemed to have been adopted best by orthoptists working in the deprived areas where many of patients spoke Dutch poorly. However, it was hard to implement the other compliance-improving measures. Although the orthoptists estimated patients' compliance to be lower after implementation (75.6% vs. 66.8%), this awareness of non-compliance was only a temporarily effect. After implementation, we did observe better communication between orthoptists and their patients. Similarly, the mean amount of time orthoptists spent on their explanations to the child had doubled, albeit by an average of less than 40 seconds; it was still only 15% of the time spent on explanations to the parent. The cartoon was made available nationwide via the department of Technology Transfer at ErasmusMC Rotterdam (a semi-governmental institution experienced in valorising medical interventions) with the support of manufacturing companies of eye patches.

Finally, **chapter 10**, discusses the results of this thesis are discussed and considers the options for further research.

SAMENVATTING

Met een prevalentie van 3-4% is amblyopie (een 'lui oog') de meest voorkomende oogaandoening op de kinderleeftijd. Het is een verminderde gezichtsscherpte in één van beide ogen welke veroorzaakt wordt door een onderbreking van de normale visuele ontwikkeling. Door het afplakken van het goede oog met een oogpleister voor het zesde levensjaar, kan de gezichtsscherpte van het amblyope oog zich herstellen. Het succes van deze behandeling is afhankelijk van verschillende factoren, zoals vroege opsporing, tijdig starten van de behandeling en goede therapietrouw van de patiënt - dat wil zeggen, de mate waarin de patiënt zich houdt aan het voorgeschreven behandeladvies van de zorgverlener.

Elektronisch gemeten therapietrouw bij de afplakbehandeling, geobjectiveerd met de Occlusion Dose Monitor (ODM), is matig: gemiddeld werd maar 57% van de voorgeschreven uren daadwerkelijk geplakt. In Nederland is in een gerandomiseerde effectiviteitstudie aangetoond dat de therapietrouw sterk verbetert bij het gebruik van een educatief programma ("*Het Plakboek*") dat ontworpen is door José Vingerling en Gerard de Bruyne. Dit programma bestaat uit een tekstloos stripverhaal dat een vierjarig kind uitlegt waarom hij/zij de oogpleister moet dragen, een kalender met beloningsstickers, en voor de ouders een informatiefolder over de aandoening dat in zes talen beschikbaar was.

Op basis van deze kennis worden de volgende doelen in dit proefschrift besproken (**hoofdstuk 1**):

- het bepalen van de bijdrage van het oogheelkundige screeningsprogramma in Nederland aan vroegtijdige opsporing van amblyopie;
- het exploreren van complexe factoren omtrent therapieontrouw die de effectiviteit van de amblyopie behandeling mogelijk beïnvloeden;
- het evalueren van de implementatie (invoering) van het educatieve programma in de dagelijkse praktijk - omdat eerder is aangetoond dat een effectief gebleken product niet altijd gemakkelijk en spontaan wordt ingevoerd in de dagelijkse praktijk.

De bijdrage van de oogheelkundige screening in Nederland op de vroegtijdige opsporing van amblyopie werd geëvalueerd in de Rotterdam AMblyopia Effectiveness Study (RAMSES). **Hoofdstuk 2** beschrijft de klinische resultaten van deze follow-up (vervolg) studie: scheelziens- en combinatieamblyopen waren gemiddeld 2,5 jaar toen zij voor het eerst een bezoek aan de orthoptist of oogarts brachten, dat is twee jaar jonger dan kinderen met een refractie amblyopie (amblyopie door ongelijke brilsterkte, 4,5 jaar). In vergelijking met een cohort die geen screening hebben ondergaan, werden deze verschillende soorten amblyopie meer dan twee jaar later gedetecteerd. Op basis van de resultaten van de

RAMSES, veronderstellen wij dat screening vanaf drie jaar, waarbij de gezichtsscherpte wordt gemeten, het meest bijdraagt aan de detectie van amblyopie. Een kwart van de kinderen met amblyopie genoot echter niet direct van het screeningsprogramma door zelfverwijzing naar de oogarts/orthoptist of door mislukte verwijzing van het consultatiebureau naar het ziekenhuis. Wij ondervonden dat mislukte verwijzingen na een positieve screening test waren veroorzaakt door ontrouw van de ouders in het opvolgen van de verwijzing, miscommunicatie tussen jeugdarts en de ouders, gebrekkige documentatie en het niet opvolgen van het verwijzingsprotocol door de jeugdarts (**hoofdstuk 3**).

Omdat therapieontrouw een belangrijke beperkende factor is voor het leveren van effectieve zorg, is het interessant om te weten hoe de behandelaars (orthoptisten) omgaan met therapieontrouw. In **hoofdstuk 4** worden de verschillen in houding en benaderingen door orthoptisten bij de therapieontrouw van patiënten besproken. Dit onderzoek leverde drie belangrijke bevindingen op. Ten eerste onderschatten orthoptisten therapieontrouw bij de afplakbehandeling. Ten tweede maakten zij gebruik van diverse praktijkgerichte oplossingen om therapieontrouw tegen te gaan. Ten derde, ook al komt therapieontrouw vooral voor bij kinderen van ouders die slecht Nederlands spreken, in achterstandswijken werd er minder tijd besteed aan uitleg dan elders in Nederland waar taalbarrières minder vaak voorkwamen. De geplande tijd voor het eerste bezoek van een nieuwe patiënt was gemiddeld 7,5 minuten korter in achterstandswijken dan in andere wijken.

Daarnaast zijn enkele determinanten van therapieontrouw bij de afplakbehandeling nog niet geïdentificeerd. In **hoofdstuk 5** hebben wij onderzocht of therapietrouw beïnvloed wordt door verschillende materiaaleigenschappen van de vier meest gebruikte merken oogpleisters: 3M (Opticlude), Master-Aid (Ortopad), Lohmann-Rauscher (Pro-ophtha) en BSN Medical (Coverlet S), en het draagcomfort van deze pleisters. Het comfort van ieder merk oogpleister was matig. Hoewel er geen verschil werd gevonden in elektronisch gemeten therapietrouw tussen de verschillende merken pleisters, werden er grote verschillen gevonden met betrekking tot de verwijdering van de pleister, de vormgeving, de lichtdoorlaatbaarheid, en de plakkracht op de huid. Met deze resultaten in achtnemend zou de keuze van het merk oogpleister op basis van de duur van het plakvoorschrift per dag gebaseerd kunnen worden.

Eerder was aangetoond dat de therapietrouw bij de afplakbehandeling bijzonder laag is bij kinderen van allochtone ouders die de Nederlandse taal slecht spreken of een laag opleidingsniveau hebben. In **hoofdstuk 6** werd bepaald welke migrantfactoren de mate van therapietrouw beïnvloeden. Wij vonden dat lage therapietrouw geassocieerd is met een hoge mate van sociale cohesie op microniveau (familie, buren en vrienden) en een lage mate van sociale cohesie op macroniveau (de Nederlandse samenleving).

Het laatste deel van dit proefschrift richt zich vooral op de implementatie van het educatieve programma en andere therapietrouwverbeterende maatregelen in de dagelijkse praktijk. **Hoofdstuk 7** beschrijft de klinische resultaten van deze implementatie. Bij kinderen uit achterstandswijken verbeterden de elektronisch gemeten therapietrouw en het opvolgen van poliklinische afspraken na implementatie van het educatieve programma, meer dan bij kinderen uit andere wijken in Nederland. De therapietrouw was gemiddeld 52% vóór de implementatie, en 62,3% nadien. De opkomst op afspraken was 60,3% vóór de implementatie, en 76% daarna. Na de implementatie verbeterde de gezichtscherpte in het amblyope oog ("Speed of Reduction in Intra-ocular Acuity Difference", SRIAD) sneller bij kinderen uit achterstandswijken dan bij kinderen uit andere wijken in Nederland. In de gehele studiepopulatie daalde het verschil in gezichtsscherpte tussen beide ogen met 2,4 regels gezichtscherpte per jaar in de groep vóór de implementatie, en met 3,0 regels gezichtscherpte per jaar in de groep na de implementatie. Omdat deze bevindingen meer uitgesproken waren in achterstandswijken, ondersteunt dit het argument dat dit programma vooral ingezet dient te worden in dergelijke wijken.
Doordat dit educatieve programma uit drie elementen bestaat (het stripverhaal, de kalender met beloningsstickers en de informatiefolder in zes talen voor de ouders) blijft het onduidelijk door welk element de therapietrouw verbetert. In een nieuwe prospectieve gerandomiseerde studie (**hoofdstuk 8**) maten we de beste elektronisch gemeten therapietrouw in de groep die het stripverhaal kreeg (89%, gemiddeld). De op een na beste was de groep die de informatiefolder kreeg (73%, gemiddeld), en de derde was de groep die de kalender met beloning stickers kreeg (67%, gemiddeld). De laagste therapietrouw was gemeten in de controlegroep, waarbij kinderen een kleurplaat ontvingen (55%, gemiddeld). In **hoofdstuk 9** wordt beschreven dat het educatieve programma wel geïmplementeerd kan worden in de dagelijkse praktijk. De beste adoptie was gemeten bij orthoptisten werkzaam in de achterstandswijken waar veel patiënten slecht Nederlands spraken. Echter, de andere therapietrouwverbeterende maatregelen waren nauwelijks geïmplementeerd. Hoewel de orthoptisten de therapietrouw bij hun patiënten lager schatten na de implementatie (75,6% versus 66,8%), was dit besef van therapieontrouw slechts een tijdelijk effect. Na de implementatie, observeerden wij wel een betere communicatie tussen orthoptist en hun patiënten. Ook was de lengte dat orthoptisten besteedden aan de uitleg aan het kind verdubbeld; zij het met een gemiddelde van minder dan 40 seconden, was het nog steeds slechts 15% van de tijd besteed aan uitleg aan de ouder. Het educatieve programma werd landelijk ter beschikking gesteld via de afdeling Kennistransfer op het ErasmusMC Rotterdam (die ervaring heeft met de valorisatie van medische interventies) met de steun van pleisterfabrikanten.

Tot slot, in **hoofdstuk 10** worden de resultaten van dit proefschrift besproken en de mogelijkheden voor verder onderzoek beschouwd.

Dankwoord

DANKWOORD

Een proefschrift zal nooit tot stand komen zonder hulp, steun, en werk van vele andere personen. En dat geldt ook voor dit proefschrift. Het uitspreken van mijn persoonlijke dank aan alle personen die nauw betrokken zijn geweest bij mijn onderzoek, die mij hulp hebben geboden bij het tot stand komen van dit proefschrift, en die voor afleiding hebben gezorgd op de momenten dat ik dat nodig had, is het minst wat ik kan doen. Dus bij dezen!

Uiteraard wil ik bedanken: promotor en begeleider, Prof.dr. H.J. Simonsz. **Huib**, zonder jou was ik nooit begonnen aan dit enorme project. Het heeft ons beiden bloed, zweet en tranen gekost, maar met dit proefschrift als resultaat. Het werken met en onder jou was een ware 'eyeopener'. Het was een onvergetelijke levenservaring, en de kennis die ik bij je heb opgedaan zal mij vast ten goede komen.

Dank gaat ook uit naar mijn andere promotor, **Prof.dr. G. van Rij**. Uw interesse en raad die boden mij steun. Uw persoonlijke betrokkenheid betreffende mij en het onderzoek heb ik altijd gewaardeerd. Dank dat u mij de mogelijkheid heeft geboden om op uw afdeling binnen het Erasmus MC te promoveren, ik heb het als een voorrecht ervaren.

Prof.dr. Vingerling, Hans, bedankt voor je deelname aan de leescommissie, maar bovenal bedankt voor je tijd, interesse, hulp en tips die je mij in het afgelopen jaar hebt gegeven.

De overige leden van de leescommissie, **Dr. Essink-Bot** en **Dr. Heerdink** bedank ik voor hun waardevolle tijd en moeite om dit manuscript in een zeer rap tempo te beoordelen. De hooggeleerde heren **Prof.dr. Passchier**, **Prof.dr. De Koning**, **Prof.dr. Schalij-Delfos** en **Dr. Juttmann** dank ik voor de bereidheid zitting te nemen in mijn promotiecommissie.

Dank gaat uit naar **Paul Heeman**, **Ruud Voorn** en **Hugo Molenaar** (afdeling MTO van het AMC in Amsterdam) zonder jullie was het ons niet gelukt om opnieuw de therapietrouw bij de plakbehandeling van kinderen met een lui oog te meten.

Ik dank **Mw. Van Rij-de Jong**, **Dhr. Alexander** en **Mw. Verhoef** voor hun hulp: de stukken zijn stukken beter uit de verf gekomen dankzij jullie.

Bijzondere dank ben ik verschuldigd aan de enorme inzet en werk van de orthoptisten die intensief hebben deelgenomen aan dit onderzoek (in willekeurige volgorde): **Karin Croft**, **Chantal Klerks - Kuijpers**, **Patricia Neomagus**, **Corlien Stoop**, **Elly van Rooijen – Troost**, **Lianne Bakker**, **Natasja Bos - De Bruin**, **Ariënne Wellner - Fokker**, **Willemijn Jutte**, **Anita Hupsch - Van den Heuvel**, **Annemieke Spaan**, **Moniek de Rooij**, **Arianne van Voorden -**

Van der Knaap, **Ellen Cockx – Huitema**, **Wanda Hoogeveen**, **Ineke Huisman**, **Ellen van Minderhout**, **Brigitte Simonsz - Tóth**, **Marleen Vermeulen - Jongen**, **Anouschka van de Bovenkamp - Scheper**, **Anke Versteeg**, **Trudie Kooij - 't Spijker**, **Marlien Kruiswijk**, **Daniëlle van Gemert**, **Arieneke Burger - Meywaard**, **Daniëlle Hamers**, **Ingrid 't Hart - Leemhuis**, **Marit Hazenkamp - Boekhold**, **Femke Jansen**, **Diet van der Nagel – Pronk**, **Sylvia Rousse**, **Dorien Valster**, en **Jacoline Kieviet - De Geus**. Ook dank dat ik bij jullie 'in de keuken' heb mogen kijken. De oogartsen en managers van de deelnemende ziekenhuizen dank ik voor de mogelijkheid om op hun afdeling onderzoek te mogen doen.

Ik dank ook alle (anonieme) orthoptisten die ooit één of meerdere vragenlijsten van ons onderzoek hebben ingevuld. Verder wil ik mijn dank uitspreken aan alle kinderen en hun familieleden die aan het onderzoek hebben deelgenomen. De wetenschap zou nergens zijn geweest zonder de bereidwilligheid van proefpersonen zoals zij!

Hilal Akcan, **Fatma Ziylan**, **Annemarie Romers** en **Xanne Slot**, bedankt voor de inspanning die jullie hebben geleverd tijdens jullie profielstages. **Caspar Looman** en **Gerard Borsboom**, ik kon bij jullie altijd even om hulp vragen als ik weer eens een statische vraag had. Bedankt voor jullie tips, uitleg en bijdrage aan mijn manuscripten. **Marianne Sinoo** bedankt voor jouw inzet, de organisatie van de cursus en de trouwe aanwezigheid van jou tijdens de bijeenkomsten in Woerden. **Heleen van Agt**, bedankt voor je inbreng aan het begin van het onderzoek. Dank gaat ook uit aan **Luc Vercauteren**: je hebt aan het begin van de studie heel nuttig advies gegeven.

Gerard de Bruyne en **José Vingerling**, dank voor jullie bijzondere werk en voor de omslag van dit proefschrift. Ik heb grote bewondering voor wat jullie doen, maken en waar jullie voor staan! Ik zie jullie creaties nog graag terug!
Hanny Groenewoud, integriteit binnen de wetenschap heb ik vooral van jou geleerd. Ik heb het als een voorrecht ervaren om met jou aan het RAMSES onderzoek te mogen werken.
Sjoukje Loudon, mijn voorgangster van dit onderzoek. Zonder jou bestond dit onderzoek niet. Bedankt voor je adviezen en nuchtere blik op het onderzoek.
Anne Roefs, bedankt voor jouw inspanningen die je hebt geleverd. Alle uurtjes die je er extra in hebt gestopt, ben ik je zeer dankbaar voor. Ook dank voor de gezelligheid!
Elizabet Vukovic, mijn naaste collega van het begin! Wat heb ik veel met jou gelachen: om foto's, tijdens huisbezoeken, op de HU en congressen. Dank voor jouw bijdrage aan het begin van de studie. Heel veel succes met je artistieke carrière!
Gerdien Holtslag, mijn naaste collega aan het eind! Je was als een geschenk uit de hemel toen je met mij het onderzoek wilde voortzetten. Jouw kennis, daadkracht en secuurheid deden het onderzoek veel goeds. Dank.

Wijnanda Asjes, mijn naaste collega van het begin tot aan het eind! Wat had ik nou zonder jou gemoeten? Ik was de 1 en jij zorgde voor de 3! Hartelijk dank voor je kritische blik op het onderzoek, je luisterend oor, en de brainstormsessies.

Aan collega's en oud-collega's van de 16e. Allereerst **Elizabeth**, kamergenootje van 1607, de woensdag was geen woensdag als jij er niet was. Succes met alles wat je nog allemaal van plan bent. **Dominiek,** ook al was je maar voor een korte periode een kamergenoot, bedankt voor je goede tips, en succes met jouw carrière stappen. **Emine**, jouw promotie was de eerste die ik zag, en blijft me altijd bij. Bedankt voor jouw adviezen. **Jan-Roelof**, samen die eeuwenoude supertrage dinosaurus van een 'computer' delen, schept toch een band ;-) Ik ben blij voor je dat je hem nu eindelijk hebt mogen inruilen! Dank dat jij jouw orthoptische kennis en ervaring met me wilde delen. **Walter**, medeaio van de eerste jaren, dank voor de gezellige koffiepauzes. **Michael**, zoals ik had beloofd: bedankt voor je bruine fiets met blauwe sticker! (hij is helaas overleden…). **Thomas**, medeaio van het laatste jaar, arts in hart en nieren, succes met jouw weg naar een goede carrière. **Erwin**, nu ook medeaio, good luck met je onderzoek. **Wishal**, medeaio van de 21e, jij wordt zo'n briljante oogarts geloof ik. **Anna**, medeaio van na mijn tijd, jammer dat ik al weg was toen jij kwam: jij gaat het helemaal maken. **Magda**, dank voor je interesse en tips! **Esmay**, ik wens je al het beste met jouw promotieonderzoek bij Huib, we'll keep in touch. **Maartje**, bedankt voor je hulp bij mijn keuzeonderzoek. **Marieke**, succes met jouw promotieonderzoek in het buitenland. **Frans-Willem**, bedankt voor de leerzame momenten tijdens de OVAS-meetings. **Sander**, de manier hoe jij als ingenieur de wetenschap interpreteert deed mijn ogen doen openen. Bedankt voor je hulp met de technische programma's en de gezelligheid op de ARVO.

Ik wil graag alle **assistenten, oud-assistenten en stafleden van de poli oogheelkunde van het Erasmus MC** enorm bedanken voor de interesse en hun vragen betreft het onderzoek. Even 'crashen' in de veilige assistentenkamer deed me altijd goed. Ik heb me altijd op mijn plek gevoeld bij jullie, en ik zal dat waarschijnlijk ook altijd houden. Ook aan al het personeel van de poli oogheelkunde, ik beloof jullie, jullie zullen me niet meer als een verloren ziel in de wandelgangen van de poli zien. **Nicole (vB)** bedankt voor je altijd snelle reacties, de moeite om mij in de drukke agenda's van de profs te kunnen proppen en de kletsmomentjes. **Isa** en **Helma**, bedankt voor jullie kritische en klinische blik die jullie als orthoptist hadden op het onderzoek.

Heel veel dank en lof gaan uit naar mijn paranimfen. **Lintje**, wat hebben we elkaar toch gevonden tijdens onze onderzoekstijd: ik kon altijd bij je terecht als er iets was. Je begrip voor, jouw kijk op en jouw gevoel van medeleven voor de situatie gaven me veel steun. Veel dank. Ik mis de wekelijkse (soms dagelijkse) Swirls – mix ijs met alléén de rode

vruchten en pure chocolade vlokken. Mochten we ooit weer samen in eenzelfde ziekenhuis werken, dan pakken we de Swirlpauzes weer op!

Jolanda, als we wilden, konden wij UREN met elkaar klessebessen, alleen zou dat niet echt verantwoord zijn geweest. Jij bent mijn gezelligste kamergenoot geweest tijdens mijn promotietijd. Ik kwam daarom ook graag op 1610 zitten. Je was vrijwel altijd het eerste aanspreekpunt in de ochtend: onder genot van een kopje koffie bespraken we dan over de normale gebeurtenissen van het leven. Het bracht me dan de beide benen weer op de grond. Jammer dat de tijd nu dan echt is aangebroken dat ik wegga. Ik hoop dat we elkaar nog vaak zullen spreken. Bedankt!

Naast alle collega's, oud-collega's en onderzoekers, ben ik dank verschuldigd aan mijn vrienden die ik de afgelopen jaren toch wel een beetje erg heb verwaarloosd… Ook aan de vrienden die ik hieronder niet persoonlijk bij naam noem! Aan mijn dispuutgenoten: ik ben nu weer in voor een biertje! Jullie? **Iziss**-promovenda, maak me trots! Lieve **Sien**, bedankt voor je geduld, zelfs na al die jaren. Dat we elkaar steeds minder zijn gaan zien, heeft onze beste vriendschap gelukkig niet geschaad. Lieve **Plien**, na 23 jaar ben je nog steeds een vriendin waar ik veel aan heb. Zelfs tijdens mijn onderzoek: sommige vraagstukken werden mij helderder na een gesprek met jou, dank!

Uiteraard veel dank aan mijn allerliefste vriendinnetjes van mijn club T: **Floor**, **Ju**, **Kar**, **Lee**, **Liek**, **Frouk**, **Nino**, en **Vi**!!! Jullie hebben mij voor zoveel gezonde en gepaste afleiding gezorgd. En jullie hebben altijd met veel interesse geluisterd en mij waardevolle tips gegevens. Het is een onbeschrijfelijk goed gevoel om te weten dat jullie, als goede vriendinnen, altijd voor me klaar staan. Ik mis jullie daarom, doordat ik jullie de afgelopen tijd zoveel minder heb kunnen zien dan dat ik oprecht had gewild. Ik hoop dat ik nog heel veel jaren samen met jullie (en de nieuwe mini-schatjes onder ons) van het leven kan genieten met lekkere wijntjes en etentjes, tijdens stapavonden en vakanties, hier en overzee.

Mijn **gehele familie** - ooms, tantes, neven, en nichten – en natuurlijk mijn schoonfamilie (in het bijzonder **Trudy** en **Jos**) dank ik voor hun liefde en interesse in de voortgang van mijn onderzoek! **Tante Ien** en **Oom Ting** bedankt voor jullie luisterend oor en tips.

In het bijzonder wil ik mijn zus, **Ier**, bedanken. Je hebt me vreselijk veel geholpen met mijn artikelen. Jij kon mij de allerbeste inhoudelijke raad en adviezen gegeven. Daar tipte niemand aan. Jammer dat jij nooit het laatste woord had. Heel veel dank voor dat je mijn grote zus wilde zijn. Ik ga er vanuit dat we die bucket list aan trekkings gaan afwerken ;-). Ik wens je veel succes met jouw promotieonderzoek en carrière! Jij gaat het helemaal maken!

Bijzonder veel dank gaat naar mijn ouders. **Ma**, dank voor je praktische inzichten, je oplossings- en organisatievermogen, dat is iets wat ik van jou heb en goed heb kunnen toepassen in dit onderzoek! Ook bedankt voor je kilo's lekkere eten: een goed gevulde diepvries was een goed begin van een week met lange onderzoeksdagen. **Pa**, ik ben blij met jou: Jij hebt me op veel facetten aangestuurd en begeleid. Je hebt me geleerd om zaken te doen, en juist precies op de moeilijkste en vervelendste momenten. Dat had ik niet van een ander kunnen en willen leren. Ik hoop nog heel veel van je te mogen leren, want er valt volgens mij nog heel wat te leren over het reilen en zeilen binnen de medische wereld…

Mijn grootste dank ben ik verschuldigd aan de persoon die het meest van mijn 'moods' heeft moeten incasseren: **Maarten**. Ze noemen personen zoals jij ook wel 'de rots in de branding': kazige tekst, maar het is de waarheid. Het moge namelijk duidelijk zijn dat jij de rots was en ik de ruwe branding! Jij bracht structuur en rust in mijn chaos. Ik heb je lief, en ik dank je voor dat je mijn leven op alle bedenkbare manieren zoveel makkelijker hebt gemaakt, voor jouw liefde, jouw humor en jouw onvoorwaardelijke steun. Door dik en dun! Ik sluit deze periode nu hier af, en hoop hierbij een start te maken waarbij wij saampies aan een mooi, nieuw, en vooral leuk avontuur gaan beginnen als twee expats in Shanghai. Heb er zin an!!

Angela

About the author & PhD Portfolio

ABOUT THE AUTHOR

Angela Mei-Lian Tjiam was born on March 1st, 1984, in Goes, The Netherlands. She attended the St. Willibrord College in Goes and graduated in 2002. In the same year, she started Medical School at the Erasmus University Rotterdam. During her study, she worked as a medical student nurse in the Havenziekenhuis (Port Hospital) in Rotterdam, and as a transport employee in the Nursing home Ter Valcke in Goes. She was also an active member of the student's association S.S.R. Rotterdam. In 2006, she started her research project on *Effective Detection and Treatment of Amblyopia: Addressing Noncompliance* at the Department of Ophthalmology at the ErasmusMC, Rotterdam, The Netherlands, under supervision of Prof.dr. G. van Rij and Prof.dr. H.J. Simonsz. After obtaining her pre-clinical medical degree in August 2006, she continued as researcher on this project. She presented her work at several national and international meetings. In October 2010, she started her clinical internships to obtain her medical degree. In July 2012, she will spend three months at the general hospital in Usangi, Tanzania, as an intern in Tropical Health. She will become a medical doctor in November 2012. Subsequently, she will move to Shanghai for several years to work as a physician in an international medical clinic. When she returns to The Netherlands, she will start her residency to become an ophthalmologist.

Erasmus MC
Universitair Medisch Centrum Rotterdam

PhD Portfolio Summary of Ms. Angela M. Tjiam

Summary of PhD training and teaching activities

Name PhD student: Angela Mei-Lian Tjiam
PhD period: 2006 ~ 2010
Erasmus MC Department: Ophthalmology
Promotors: Prof.dr. G. van Rij
Prof.dr. H.J. Simonsz

1. PhD training	**Year**	**Workload**
General academic skills		
a) Biomedical English Writing and Communication (Erasmus MC)	2006~2007	3.0 ECTS
b) Academic Writing for PHD students (EUR)	2009	18.5 Hrs
Research skills		
a) A First Glance at SPSS for Windows (SC05)	2008	1.55 ECTS
b) Classical Methods for Data-analysis (CC02)	2008	5.7 ECTS
c) CPO Minicursus "Methodologie van Patiëntgebonden Onderzoek en Voorbereiding van Subsidieaanvragen"	2010	8 Hrs
Presentations on conferences and seminars		
a) Association for Research in Vision and Ophthalmology (ARVO) annual meeting, USA; poster presentations	2006~2009	4.0 ECTS
b) European Symposium on PAtient COMpliance and Persistence (ESPACOMP) annual meeting, D, F, CH; 1 oral & 2 poster presentations	2006~2008	3.5 ECTS
j) Weekly ophthalmology meetings, ErasmusMC Rotterdam, NL; oral presentations	2006, 2009	2.0 ECTS
c) Nederlands Oogheelkundig Gezelschap (NOG) annual meeting, NL; 3 oral & 1 poster presentations	2007~2009	4.0 ECTS
d) Rotterdam Amblyopia Meeting (RAM), NL; oral presentations	2007, 2009	2.0 ECTS
e) Nederlands Vereniging van Orthoptisten (NVvO) biannual meeting, NL; oral presentations	2007, 2009	2.0 ECTS
f) Child Vision and Research Society (CVRS) meeting, GB; oral presentation	2007	1.5 ECTS
g) Optometristen Vereniging Nederland annual meeting (OVN), NL; oral presentation	2007	1.0 ECTS
h) Bielschowsky Gesellschaft, D; poster presentation	2007	1.5 ECTS
i) International Orthoptic Association (IOA) conference, B; poster presentations	2008	1.0 ECTS
(Inter)national conferences, seminars and workshops		
a) Kennis Beter Delen conference, NL	2008	16 Hrs
b) Tweede Nationale Therapietrouw conference, NL	2008	8 Hrs
c) NOG annual meeting, NL	2010	8 Hrs
d) Donders Gezelschap, NL	2006~2008	12 Hrs
e) NVvO biannual meeting, NL	2007~2009	12 Hrs
f) Workshop: Mind-mapping, Fast-reading, Brain-training, EUR, NL	2009	20 Hrs

2. Teaching activities	Year	Workload
Lecturing		
a) Training course: Compliance in the orthoptic practice, University of Applied Sciences Utrecht, dept. of Orthoptics, NL	2007(2x)	4.0 ETCS
b) Biannual meetings of participants and biannual meetings of study committee of the Implementation study of a compliance-improving measure for amblyopia treatment	2006~2010	128 Hrs
Supervision		
a) Supervising Orthoptic students of University of Applied Sciences Utrecht (2 students)		
Thesis: A study in different religious, ethnic and cultural predictors of compliance with occlusion therapy for amblyopia	2008	64 Hrs
b) Supervising Orthoptic students of University of Applied Sciences Utrecht (2 students)		
Thesis: The degree of identification of symbols of Landolt-C, E-symbols, Lea Hyvärinen, ETDRS and the Amsterdam Picture Chart by young adults: a randomised comparison study	2008~2009	64 Hrs
c) Assisting supervision Master Biomedical Engineering student of TU Delft with assignment for Strabismus Surgery course (1 student)		
Report: ODM-analysis	2008	4 Hrs
Other		
a) Developing and organising a 3-day training course: Compliance in the orthoptic practice, in cooperation with dept. of Orthoptics, University of Applied Sciences Utrecht, NL	2007 (2x)	200 Hrs
TOTAL Workload		**36.75 ECTS & 562,5 Hours**

Appendices

APPENDICES

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Appendix II) The Dutch Screening Programme (Chapter 2)

Appendix III) Referral letter, RAMSES (Appendix II, Chapter 2)

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Appendix X) The educational cartoon for wearing glasses (Chapter 9)

Appendix XI) Data collection and statistical analysis of Chapter 9

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Appendix I) Referral criteria for preverbal and preschool vision screening in the Netherlands (Chapter 2 and 3)

Referral and recall criteria for preverbal vision screening according to the 2002 guidelines for the detection of visual disorders in children aged 0-19 (Van Velzen-Mol 2002)

Screening test, age	**Referral criteria**	**Recall † criteria** The test should be repeated within six weeks. If the test result is the same or worse, the child still should be referred.
VOV-test*, 9, 14 and 24 months	if one or more items are abnormal	if one or more items are doubtful

* VOV = Vroegtijdige Onderkenning Visuele stoornissen, or Early Detection of Visual Disorders (i.e., inspection of the eye, pupil and pupillary reaction, corneal light reflex test, the cover-uncover test, an examination of ocular pursuit movements, and the pupillary light reflex test)

Referral and recall criteria for preschool vision screening according to the 2002 guidelines for the detection of visual disorders in children aged 0-19 (Van Velzen-Mol 2002)

Screening test, age	**Referral criteria**	**Recall † criteria** The test should be repeated within three months. If the test result is the same or worse, the child still should be referred.
APK*, 36 months	VA ≤ 5/15 in either eye Difference of ≥ 2 lines	VA of 5/10 or 5/6 in the worse eye, with an intraocular difference of no more than one line
Landolt-C, 45 months	VA ≥0.3 logMAR in either eye Difference of > 2 lines	difference of 2 lines if VA is <0.3 logMAR in both eyes
APK, 45 months (if Landolt-C fails)	VA ≤ 5/6 in either eye Difference of ≥ 2 lines	VA of 5/5 in the worse eye, with an intraocular difference of no more than one line
Landolt-C, 60-72 months	VA ≥0.3 logMAR in either eye Difference of ≥ 2 lines	VA of 0.2 logMAR in the worse eye, with an intraocular difference of no more than one line
	In Rotterdam, these criteria are also applied for VA measurements using Landolt-C at age 54 months.	In Rotterdam, the stereoacuity is done in such cases; if the stereoacuity is positive, the child passes the screening (i.e., a negative screening result)

* APK = Amsterdam Picture Chart. 5/5 was considered to be equivalent to 0.0 logMAR

Appendix II) The Dutch Childhood Vision Screening Programme (Chapter 3)

In the Netherlands, childhood vision screening consists of preverbal vision screening at 1-2, 3-4, 6-9, 14, and 24 months (examinations at 14 and 24 months were recently joined), and of preschool vision screening at 36, 45-54, and 60-72 months (Van Velzen-Mol 2002). Preverbal screening includes a corneal light reflex test, the cover-uncover test, an examination of ocular pursuit movements for both monocular and binocular conditions, inspection of the eyelids and anterior segment of the eye (particularly the cornea), inspection of the colour and shape of the pupil, and the pupillary light reflex test. Preschool screening consists of inspection, testing of monocular visual acuity, ocular alignment, and stereoacuity. At age 36 months, acuity is tested by means of the Amsterdam Picture Chart (Amsterdamse Plaatjeskaart, or APK). This chart is not logarithmic and does not use standardized optotypes. From the age of 45 months, acuity is measured by means of the Landolt-C chart. The APK is used instead if the child does not seem to understand Landolt-C testing. Vision screening is performed at child healthcare centres and public health services by specially trained screening nurses and physicians (Chapter 2).

The referral and recall criteria of the vision screening tests are presented in Appendix I. If an outcome is doubtful because the child was, for example, uncooperative at that time, the test should be repeated within 6 weeks for preverbal screening and after 3 months for preschool screening. If the test outcome is abnormal, (i.e., the screening test is positive) the child is referred to an orthoptist or an ophthalmologist. This course of referral from screening physician to ophthalmologist or orthoptist usually goes via the general practitioner (GP) who has the official gate-keeper role for most other healthcare services in the Netherlands. When a referral is made, the parents will receive a letter referring them to their GP requesting him/her to refer to an ophthalmologist or orthoptist (Appendix III). This letter contains the results of the screening test. The GP may choose to re-examine the child's vision before making a further referral to an ophthalmology department. Nearly all orthoptists working in the Netherlands practice at ophthalmology departments in hospitals (Chapter 2).

Appendix III) Referral letter, RAMSES (Appendix II, Chapter 2)

To the General Practitioner of:	**From the Child Healthcare Centre (CHC):**
Surname:	CHC Address:
First Name:	CHC Physician:
Date of Birth:	**Contact Information of General Practitioner.**
Name:	Name:
Client No.:	Address:

Dear Colleague,

This child is a participant in the project called, "The results and costs of amblyopia prevention in child health care". A screening performed during this project has resulted in the following findings.

VOV*		Date of 1st Examination :		Date of 2nd Examination :		Visual Acuity APK†	
		R	**L**	**R**	**L**	Date #1 examination	
Light Reflex Test:	Symmetric (+) / Not Symmetric (-)						
Cover Test:	No Movement (+) / Deviation With Eye Movement **During** Cover-Uncover Test (-) No Deviation (+) / Manifest Deviation **After** Cover Uncover Test (-)					Right	5/
						Left	5/
Eye Motility:	Monocular: Smooth (+) / Saccadic (-)					Date #2 examination	
	Binocular: Smooth (+) / Saccadic (-)						
Cornea:	Normal (+) / Abnormal (-)					Right	5/
Pupil:	Black (+) / Not Black (-)					Left	5/
	Round (+) / Not Round (-)						
Pupillary Reaction:	Rapid (+) / Not Rapid (-)						

This is an abnormal screening test result which was performed for the first time / subsequent time.
You are kindly requested to complete the form at the bottom of this letter and to return this letter in its entirety by using the self-addressed, stamped envelope enclosed. The attached copy may be necessary for the parents to hand over to the ophthalmologist.

Sincerely,

⓪ After having reviewed the test results, I hereby refer this patient to the Ophthalmology Department of the following hospital: .....
⓪ I have decided to decline referring this patient,
because the patient is already under treatment by the Ophthalmologist at the following hospital: ……

Signed, ..., General Practitioner Date

Appendix IV) The geographical map of The Netherlands (country) and the locations of the participating clinics (Chapter 4, 7 and 9)

Each dot represents a clinic. Black dots are clinics in low-SES areas; gray-black dots are clinics elsewhere in The Netherlands (NL). The table depicts the average [range] population, proportion of non-natives, mean income per resident, and social security welfare payment (Statistics Netherlands Accessed on Feb 26, 2010) in The Netherlands and the areas where clinics of both groups were located.

	Population	Proportion non-natives	Mean income per resident	Social security welfare payment
		%	€1 000	per 1 000 households
The Netherlands	16 305 530	19	12.2	52
Low-SES	17 882 [31 200 – 148 80]	50 [21 – 89]	10.6 [7.2 – 12.2]	139 [59 – 282]
Elsewhere in NL	90 151 [8 300 – 208 460]	19 [4 – 34]	12.3 [11.0 – 14.0]	48 [15 – 87]

Appendix V) Results of the extended structured questionnaire for participating orthoptists (Chapter 4)

Appendix V) The extended structured questionnaire for orthoptists in both groups. Group A represents the orthoptists in non-native, low-SES areas, Group B represents those working elsewhere in the Netherlands. It contained 125 questions in two parts: part I, "Orthoptists' individual personality features," and part II, "Attitude towards non compliance."

Part I) "Orthoptists' individual features"	Group A (n = 9) N (%)						Group B (n = 23) N (%)						
	(Almost) always	Often	Quite often	Sometimes	Occasionally	(Almost) never	(Almost) always	Often	Quite often	Sometimes	Occasionally	(Almost) never	P-value
Autonomy													
At work, I make decisions on my own	5 (55.6)	4 (44.4)	-	-	-	-	18 (78.3)	5 (21.7)	-	-	-	-	0.206
I know how to tackle problems	3 (33.3)	3 (33.3)	3 (33.3)	-	-	-	8 (34.8)	10 (43.5)	4 (17.4)	1 (4.3)	-	-	0.739
I work without the support of others	3 (33.3)	4 (44.4)	2 (22.2)	-	-	-	11 (47.8)	7 (30.4)	1 (4.3)	2 (8.7)	2 (8.7)	-	0.720
I know exactly what I need to do	3 (33.3)	4 (44.4)	2 (22.2)	-	-	-	4 (17.4)	15 (65.2)	4 (17.4)	-	-	-	0.651
I do not ask for advice before I make a decision	-	3 (33.3)	6 (66.7)	-	-	-	5 (21.7)	14 (60.9)	-	-	4 (17.4)	-	0.511
I do not have a 'wait-and see' attitude	-	3 (33.3)	2 (22.2)	3 (33.3)	1 (11.1)	-	-	10 (43.5)	2 (8.7)	2 (8.7)	9 (39.1)	-	0.374
I find it easy to make choices	-	1 (11.1)	4 (44.4)	3 (33.3)	1 (11.1)	-	-	8 (34.8)	11 (47.8)	2 (8.7)	1 (4.3)	1 (4.3)	0.404
I do not doubt the diagnoses I make	-	3 (33.3)	4 (44.4)	2 (22.2)	-	-	3 (13.0)	9 (39.1)	2 (8.7)	-	9 (39.1)	-	0.109
Organizational skills													
I am precise in my work	2 (22.2)	4 (44.4)	2 (22.2)	1 (11.1)	-	-	6 (26.1)	12 (52.2)	5 (21.7)	-	-	-	0.510
I manage my workload adequately	3 (33.3)	5 (55.6)	1 (11.1)	-	-	-	8 (34.8)	11 (47.8)	4 (17.4)	-	-	-	0.890
I work carefully	6 (66.7)	2 (22.2)	1 (11.1)	-	-	-	9 (39.1)	12 (52.2)	2 (8.7)	-	-	-	0.255
I finish everything I start	5 (55.6)	4 (44.4)	-	-	-	-	7 (30.4)	14 (60.9)	2 (8.7)	-	-	-	0.152
I comply with agreements made	6 (66.7)	3 (33.3)	-	-	-	-	15 (65.2)	8 (34.8)	-	-	-	-	0.939
I stay within the time period allotted per visit	2 (22.2)	3 (33.3)	4 (44.4)	-	-	-	4 (17.4)	11 (47.8)	5 (21.7)	3 (13.0)	-	-	0.965

My work often proceeds according to plan	1 (11.1)	5 (55.6)	2 (22.2)	1 (11.1)	-	-	2 (8.7)	9 (39.1)	7 (30.4)	3 (13.0)	2 (8.7)	-	0.340
It requires no effort on my part to keep my work area nice and tidy	2 (22.2)	4 (44.4)	1 (11.1)	1 (11.1)	-	1 (11.1)	8 (34.8)	6 (26.1)	6 (26.1)	1 (4.3)	2 (8.7)	-	0.696
I'm not untidy at work	3 (33.3)	5 (55.6)	1 (11.1)	-	-	-	11 (47.8)	7 (30.4)	3 (13.0)	2 (8.7)	-	-	0.821
I sometimes misplace things at work	-	-	1 (11.1)	2 (22.2)	3 (33.3)	3 (33.3)	-	1 (4.3)	-	4 (17.4)	9 (39.1)	9 (39.1)	0.578
If something comes to mind, I need to do it immediately, otherwise I'll forget to do it	-	2 (22.2)	2 (22.2)	5 (55.6)	-	-	1 (4.3)	6 (26.1)	2 (8.7)	11 (47.8)	3 (13.0)	-	0.751
Ability to comprehend													
I feel closely associated with my orthoptic colleagues	2 (22.2)	7 (77.8)	-	-	-	-	8 (34.8)	12 (52.2)	2 (8.7)	-	-	1 (4.3)	0.904
I feel involved with my patients	2 (22.2)	7 (77.8)	-	-	-	-	9 (39.1)	11 (47.8)	3 (13.0)	-	-	-	0.740
I take the patient's wishes into account	3 (33.3)	5 (55.6)	1 (11.1)	-	-	-	7 (30.4)	14 (60.9)	2 (8.7)	-	-	-	0.962
I can empathize well	4 (44.4)	5 (55.6)	-	-	-	-	9 (39.1)	12 (52.2)	1 (4.3)	-	1 (4.3)	-	0.619
I listen to the patient well	4 (44.4)	4 (44.4)	1 (11.1)	-	-	-	10 (43.5)	11 (47.8)	2 (8.7)	-	-	-	0.981
I listen to my colleagues well	4 (44.4)	5 (55.6)	-	-	-	-	8 (34.8)	13 (56.5)	2 (8.7)	-	-	-	0.474
I give the patient enough time to ask questions	4 (44.4)	5 (55.6)	-	-	-	-	14 (60.9)	8 (34.8)	1 (4.3)	-	-	-	0.484
I find it difficult to understand foreign cultures	-	-	-	5 (55.6)	2 (22.2)	2 (22.2)	-	1 (4.3)	2 (8.7)	12 (52.2)	6 (26.1)	2 (8.7)	0.323
If there is a foreign name on my consultation list, I prepare myself for a patient who does not speak the Dutch language	-	-	1 (11.1)	4 (44.4)	4 (44.4)	-	1 (4.3)	1 (4.3)	4 (17.4)	10 (43.5)	4 (17.4)	3 (13.0)	0.465

If I have a patient who speaks poor Dutch, I often notice that this person does not listen well, either	1 (11.1)	-	-	5 (55.6)	2 (22.2)	1 (11.1)	1 (4.3)	2 (8.7)	4 (17.4)	12 (52.2)	4 (17.4)	-	0.217
If I become aware that the treatment regimen I had planned is not feasible to carry out, I still keep to it	-	1 (11.1)	-	2 (22.2)	4 (44.4)	2 (22.2)	-	1 (4.3)	2 (8.7)	12 (52.2)	8 (34.8)	-	0.095
I carry out my work in a hurry	1 (11.1)	-	-	4 (44.4)	4 (44.4)	-	-	1 (4.3)	-	8 (34.8)	8 (34.8)	3 (13.0)	0.245
Flexibility													
I improvise easily	1 (11.1)	2 (22.2)	2 (22.2)	4 (44.4)	-	-	2 (8.7)	8 (34.8)	5 (21.7)	6 (26.1)	2 (8.7)	-	0.777
I cooperate pleasantly with others	4 (44.4)	2 (22.2)	3 (33.3)	-	-	-	11 (47.8)	9 (39.1)	2 (8.7)	1 (4.3)	-	-	0.571
I am fond of changes	1 (11.1)	-	1 (11.1)	4 (44.4)	3 (33.3)	-	-	1 (4.3)	2 (8.7)	13 (56.5)	4 (17.4)	3 (13.0)	0.664
If necessary, I'll take over the work of a colleague immediately	1 (11.1)	4 (44.4)	3 (33.3)	-	1 (11.1)	-	3 (13.0)	11 (47.8)	6 (26.1)	3 (13.0)	-	-	0.788
Sometimes, I find that I've taken on unnecessary work	-	-	1 (11.1)	6 (66.7)	1 (11.1)	1 (11.1)	2 (8.7)	5 (21.7)	3 (13.0)	10 (43.5)	3 (13.0)	-	0.076
At work, I have time for extra activities	-	1 (11.1)	-	7 (77.8)	1 (11.1)	-	1 (4.3)	1 (4.3)	2 (8.7)	9 (39.1)	9 (39.1)	1 (4.3)	0.254
I am not easily convinced of something	1 (11.1)	-	-	6 (66.7)	2 (22.2)	-	-	2 (8.7)	4 (17.4)	11 (47.8)	5 (21.7)	1 (4.3)	0.837
Early adopter													
I keep my level of expertise current	2 (22.2)	5 (55.6)	1 (11.1)	1 (11.1)	-	-	6 (26.1)	12 (52.2)	3 (13.0)	1 (4.3)	1 (4.3)	-	0.982
I participate actively in the NVvO (Orthoptic Association)	-	-	-	3 (33.3)	4 (44.4)	2 (22.2)	2 (8.7)	5 (21.7)	2 (8.7)	10 (43.5)	1 (4.3)	3 (13.0)	0.009*
I encourage others to implement innovative ideas	-			5 (55.6)	2 (22.2)	-	1 (4.3)	-	6 (26.1)	8 (34.8)	8 (34.8)	-	0.807

I participate, or would be willing to participate, in NVvO committees	-		-	4 (44.4)	1 (11.1)	3 (33.3)	2 (8.7)	3 (13.0)	2 (8.7)	6 (26.1)	5 (21.7)	5 (21.7)	0.464
I happily cooperate with transitions	1 (11.1)	1 (11.1)	1 (11.1)	5 (55.6)	1 (11.1)	-	1 (4.3)	4 (17.4)	8 (34.8)	9 (39.1)	1 (4.3)	-	0.399
I hold back on implementing innovative ideas until I am sure it is something good	-	2 (22.2)	4 (44.4)	2 (22.2)	1 (11.1)	-	1 (4.3)	4 (17.4)	7 (30.4)	9 (39.1)	2 (8.7)	-	0.709
Hierarchy													
It is clear to me who the manager of my orthoptic colleagues is	1 (11.1)	2 (22.2)	2 (22.2)	1 (11.1)	2 (22.2)	1 (11.1)	-	7 (30.4)	2 (8.7)	4 (17.4)	8 (34.8)	2 (8.7)	0.560
The ophthalmologist treats as someone in a lower position	-	-	-	1 (11.1)	5 (55.6)	3 (33.3)	-	-	2 (8.7)	6 (26.1)	5 (21.7)	10 (43.5)	0.723
I easily accept someone else taking the lead	-	2 (22.2)	3 (33.3)	(44.4)	-	-	1 (4.3)	8 (34.8)	8 (34.8)	5 (21.7)	1 (4.3)	-	0.301
If a person of a higher rank asks me to do something, I will do it immediately	1 (11.1)	-	2 (22.2)	6 (66.7)	-	-	-	1 (4.3)	6 (26.1)	15 (65.2)	-	1 (4.3)	0.671
I consider the hierarchy within the department to be important	-	1 (11.1)	-	4 (44.4)	2 (22.2)	2 (22.2)	-	2 (8.7)	2 (8.7)	8 (34.8)	7 (30.4)	4 (17.4)	0.913
	Strongly agree	Somewhat agree	Neither agree nor disagree	Disagree somewhat	Strongly disagree		Strongly agree	Some what agree	Neither agree nor disagree	Disagree somewhat	Strongly disagree		
If I instruct a patient to do something, he / she must obey	4 (44.4)	1 (11.1)	2 (22.2)	1 (11.1)	1 (11.1)		7 (30.4)	10 (43.5)	6 (26.1)	-	-		0.708
I consider myself to be one of the doctor's assistants	6 (66.7)	1 (11.1)	1 (11.1)	1 (11.1)	-		16 (69.9)	5 (21.7)	2 (8.7)	-	-		0.682
I find it annoying when the ophthalmologist treats me like a subordinate	6 (66.7)	3 (33.3)	-	-	-		15 (65.2)	7 (30.4)	1 (4.3)	-	-		0.879

	(Almost) always	Often	Quite often	Sometimes	Occasionally	(Almost) never	(Almost) always	Often	Quite often	Sometimes	Occasionally	(Almost) never	
Assertiveness													
If I disagree with colleagues, I can tell them in a calm, reasonable manner	3 (33.3)	2 (22.2)	2 (22.2)	2 (22.2)	-	-	6 (26.1)	9 (39.1)	6 (26.1)	2 (8.7)	-	-	0.776
I compliment my colleagues when they have done something right	2 (22.2)	1 (11.1)	1 (11.1)	5 (55.6)	-	-	3 (13.0)	8 (34.8)	4 (17.4)	8 (34.8)	-	-	0.523
If colleagues have done something wrong, I can calmly tell them so	2 (22.2)	3 (33.3)	1 (11.1)	3 (33.3)	-	-	2 (8.7)	14 (60.9)	4 (17.4)	2 (8.7)	-	1 (4.3)	0.766
I can express my anger at work without losing control of it	3 (33.3)	2 (22.2)	-	3 (33.3)	1 (11.1)	-	5 (21.7)	7 (30.4)	3 (13.0)	5 (21.7)	3 (13.0)	-	0.829
I express my ideas during meetings	2 (22.2)	2 (22.2)	2 (22.2)	3 (33.3)	-	-	3 (13.0)	9 (39.1)	7 (30.4)	2 (8.7)	2 (8.7)	-	0.811
I can defend myself in difficult situations	1 (11.1)	1 (11.1)	6 (66.7)	1 (11.1)	-	-	1 (4.3)	10 (43.5)	2 (8.7)	7 (30.4)	3 (13.0)	-	0.712
I am critical towards patients	1 (11.1)	6 (66.7)	-	1 (11.1)	1 (11.1)	-	4 (17.4)	11 (47.8)	4 (17.4)	2 (8.7)	2 (8.7)	-	0.909
I immediately ask for my colleague's help when I cannot resolve something myself	2 (22.2)	-	2 (22.2)	4 (44.4)	-	-	1 (4.3)	4 (17.4)	4 (17.4)	10 (43.5)	4 (17.4)	-	0.317
If I want to direct a comment to someone, I will immediately do so	-	1 (11.1)	1 (11.1)	4 (44.4)	3 (33.3)	-	-	1 (4.3)	9 (39.1)	7 (30.4)	6 (26.1)	-	0.468
I am very critical of the work of others	1 (11.1)	2 (22.2)	5 (55.6)	1 (11.1)	-	-	2 (8.7)	8 (34.8)	9 (39.1)	2 (8.7)	2 (8.7)	-	0.929
I refuse to carry out delegated tasks that I do not wish to do	-	-	1 (11.1)	4 (44.4)	2 (22.2)	2 (22.2)	-	-	1 (4.3)	10 (43.5)	7 (30.4)	5 (21.7)	0.688
I admit my mistakes	3 (33.3)	1 (11.1)	5 (55.6)	-	-	-	4 (17.4)	9 (39.1)	8 (34.8)	2 (8.7)	-	-	0.859

I can tell a colleague if he / she is being unreasonable	-	1 (11.1)	4 (44.4)	3 (33.3)	1 (11.1)	-	-	5 (21.7)	7 (30.4)	10 (43.5)	1 (4.3)	-	0.772
I am confident in my decisions	3 (33.3)	3 (33.3)	3 (33.3)	-	-	-	4 (17.4)	14 (60.9)	4 (17.4)	1 (4.3)	-	-	0.836
I can tell colleagues that I disagree with them	2 (22.2)	2 (22.2)	4 (44.4)	1 (11.1)	-	-	2 (8.7)	10 (43.5)	6 (26.1)	5 (21.7)	-	-	0.758
I allow others to express their opinions, even when I disgree with them	2 (22.2)	6 (66.7)	1 (11.1)	-	-	-	7 (30.4)	11 (47.8)	4 (17.4)	1 (4.3)	-	-	0.945
I hesitate when I must ask the ophthalmologist a question about a patient	-	-	-	-	5 (55.6)	4 (44.4)	-	-	-	4 (17.4)	8 (34.8)	11 (47.8)	0.765
Character													
I have a good overview of the work I need to carry out	2 (22.2)	4 (44.4)	3 (33.3)	-	-	-	5 (21.7)	9 (39.1)	7 (30.4)	2 (8.7)	-	-	0.723
I become easily tensed when working and from everything associated with it	-	-	2 (22.2)	4 (44.4)	3 (33.3)		-	1 (4.3)	2 (8.7)	7 (30.4)	9 (39.1)	4 (17.4)	0.178
I gladly take the lead	1 (11.1)	1 (11.1)	2 (22.2)	4 (44.4)	1 (11.1)	-	1 (4.3)	5 (21.7)	2 (8.7)	8 (34.8)	6 (26.1)	1 (4.3)	0.421
I regularly confront patients about their behavior	1 (11.1)	-	2 (22.2)	4 (44.4)	1 (11.1)	1 (11.1)	-	2 (8.7)	4 (17.4)	12 (52.2)	5 (21.7)	-	0.857
I often say what I think	3 (33.3)	1 (11.1)	2 (22.2)	2 (22.2)	1 (11.1)	-	1 (4.3)	12 (52.2)	5 (21.7)	4 (17.4)	1 (4.3)	-	0.948
I am spontaneous	-	1 (11.1)	-	2 (22.2)	4 (44.4)	2 (22.2)	-	2 (8.7)	1 (4.3)	10 (43.5)	8 (34.8)	2 (8.7)	0.258

Part II) "Attitude towards noncompliance"	Group A (n = 9) N (%)					Group B (n = 23) N (%)					
	Strongly agree	Somewhat agree	Neither agree nor disagree	Somewhat disagree	Strongly disagree	Strongly agree	Somewhat agree	Neither agree nor disagree	Somewhat disagree	Strongly disagree	P-value
One can do little to counteract noncompliance	-	1 (11.1)	3 (33.3)	5 (55.6)	-	-	3 (13.0)	2 (8.7)	6 (26.1)	12 (52.5)	0.027*
I do believe it important to find out whether patients have carried out the therapy I prescribed	-	1 (11.1)	-	1 (11.1)	7 (77.8)	1 (4.3)	2 (8.7)	1 (4.3)	4 (17.4)	15 (65.2)	0.506
I am not worried about my patients' compliance with treatment	-	1 (11.1)	-	2 (22.2)	6 (66.7)	1 (4.3)	2 (8.7)	2 (8.7)	4 (17.4)	14 (60.9)	0.681
My patients almost always occlude as prescribed	-	2 (22.2)	2 (22.2)	2 (22.2)	3 (33.3)	5 (21.7)	12 (52.5)	3 (13.0)	3 (13.0)	-	0.003*
Thanks to my explanation, I am certain that my patients comply with the therapy as prescribed	-	1 (11.1)	2 (22.2)	4 (44.4)	2 (22.2)	1 (4.3)	14 (60.9)	4 (17.4)	3 (13.0)	1 (4.3)	0.003*
It is not my responsibility if patients do not patch as prescribed	2 (22.2)	-	2 (22.2)	4 (44.4)	1 (11.1)	1 (4.3)	7 (30.4)	-	10 (43.5)	4 (17.4)	0.645
I have no problem with parents who speak the Dutch language poorly, or not at all	2 (22.2)	4 (44.4)	-	3 (33.3)	-	3 (13.0)	5 (21.7)	6 (26.1)	6 (26.1)	2 (8.7)	0.302
Poor compliance occurs once in a while	3 (33.3)	5 (55.6)	-	-	1 (11.1)	14 (60.9)	5 (21.7)	3 (13.0)	1 (4.3)	-	0.287
I am not looking forward to changing my routine in order to improve compliance	2 (22.2)	-	1 (11.1)	3 (33.3)	3 (33.3)	-	1 (4.3)	3 (13.0)	8 (34.8)	11 (47.8)	0.272

I immediately read newly-published literature about compliance	3 (33.3)	6 (66.7)	-	-	-	4 (17.4)	11 (47.8)	7 (30.4)	1 (4.3)	-	0.062
I need more time to apply compliance-improving measures	1 (11.1)	4 (44.4)	2 (22.2)	2 (22.2)	-	2 (8.7)	6 (26.1)	7 (30.4)	5 (21.7)	3 (13.0)	0.289
My department needs extra time to apply compliance-improving measures	1 (11.1)	5 (55.6)	1 (11.1)	2 (22.2)	-	1 (4.3)	6 (26.1)	7 (30.4)	7 (30.4)	2 (8.7)	0.102
I sometimes wonder whether patients really occlude as prescribed, but often take no subsequent action	-	1 (11.1)	1 (11.1)	3 (33.3)	4 (44.4)	1 (4.3)	3 (13.0)	2 (8.7)	9 (39.1)	8 (34.8)	0.610
It sometimes happens that I cannot explain what I mean to the parent, as he / she does not speak Dutch	1 (11.1)	6 (66.7)	-	1 (11.1)	1 (11.1)	3 (13.0)	11 (47.8)	3 (13.0)	1 (4.3)	5 (21.7)	0.552
I believe that compliance is an important factor in my work	4 (44.4)	4 (44.4)	1 (11.1)	-	-	11 (47.8)	9 (39.1)	1 (4.3)	2 (8.7)	-	0.963
My colleagues and I have discussed the topic of improving compliance with occlusion therapy	2 (22.2)	1 (11.1)	2 (22.2)	-	4 (44.4)	3 (13.0)	5 (21.7)	7 (30.4)	3 (13.0)	5 (21.7)	0.666
Many of my patients tend not to occlude as prescribed	2 (22.2)	2 (22.2)	1 (11.1)	1 (11.1)	3 (33.3)	1 (4.3)	1 (4.3)	-	7 (30.4)	14 (60.9)	0.043*
I am willing to deal with noncompliance by using another approach over the short term (eg, within 2 months)	3 (33.3)	4 (44.4)	-	1 (11.1)	1 (11.1)	9 (39.1)	7 (30.4)	5 (21.7)	2 (8.7)	-	0.825

I am aware that I fail to contend with poor compliance, and I wish to do something about it soon	-	4 (44.4)	-	1 (11.1)	4 (44.4)	1 (4.3)	5 (21.7)	4 (17.4)	9 (39.1)	4 (17.4)	0.696
I would like to learn more about improving compliance as soon as possible	2 (22.2)	4 (44.4)	2 (22.2)	1 (11.1)	-	5 (21.7)	11 (47.8)	6 (26.1)	1 (4.3)	-	0.857
Dealing with patients who do not speak Dutch is a long-term problem	1 (11.1)	-	1 (11.1)	2 (22.2)	5 (55.6)	1 (4.3)	3 (13.0)	7 (30.4)	4 (17.4)	8 (34.8)	0.263
I am prepared to change my approach towards noncompliance over the short term (eg, within 2 months)	3 (33.3)	6 (66.7)	-	-	-	6 (26.1)	11 (47.8)	5 (21.7)	1 (4.3)	-	0.239
I am prepared to learn several new approaches in dealing with non-native patients over the short term (eg, within 2 months)	5 (55.6)	3 (33.3)	-	1 (11.1)	-	7 (30.4)	6 (26.1)	5 (21.7)	3 (13.0)	1 (4.3)	0.145
If I identify patient noncompliance, I use all possible means to deal with it	3 (33.3)	4 (44.4)	-	2 (22.2)	-	13 (56.5)	8 (34.8)	1 (4.3)	-	-	0.115
I am improving the techniques I use to stimulate compliance	2 (22.2)	-	2 (22.2)	2 (22.2)	3 (33.3)	1 (4.3)	5 (21.7)	7 (30.4)	8 (34.8)	2 (8.7)	0.476
I try to assist non-native patients with their treatment more now than half a year ago	1 (11.1)	1 (11.1)	-	1 (11.1)	6 (66.7)	-	4 (17.4)	7 (30.4)	6 (26.1)	6 (26.1)	0.156
When I notice noncompliance, I try to counteract it more now than half a year ago	1 (11.1)	1 (11.1)	1 (11.1)	-	6 (66.7)	1 (4.3)	5 (21.7)	7 (30.4)	7 (30.4)	3 (13.0)	0.093

My approach toward patients who are poorly fluent in the Dutch language has not changed over the past half year	6 (66.7)	-	1 (11.1)	1 (11.1)	1 (11.1)	14 (60.9)	3 (13.0)	3 (13.0)	2 (8.7)	1 (4.3)	0.981
My approach toward the patient has changed over the past six months	1 (11.1)	-	1 (11.1)	-	7 (77.8)	1 (4.3)	2 (8.7)	6 (26.1)	4 (17.4)	10 (43.5)	0.184
My opinion on non-native patients has changed over the last half year	1 (11.1)	-	2 (22.2)	-	6 (66.7)	-	1 (4.3)	2 (8.7)	9 (39.1)	11 (47.8)	0.800
My opinion on patients from low-SES areas has changed over the last half year	1 (11.1)	2 (22.2)	-	6 (66.7)	-	-	6 (26.1)	7 (30.4)	9 (39.1)	-	0.573
I consistently contend with poor compliance	2 (22.2)	6 (66.7)	1 (11.1)	-	-	6 (26.1)	9 (39.1)	5 (21.7)	1 (4.3)	1 (4.3)	0.558
Nowadays, I apply my knowledge about compliance in practice with a greater awareness than in the past	3 (33.3)	2 (22.2)	1 (11.1)	1 (11.1)	2 (22.2)	3 (13.0)	5 (21.7)	7 (30.4)	5 (21.7)	3 (13.0)	0.493
I discuss compliance issues with my colleagues on regular basis	1 (11.1)	1 (11.1)	2 (22.2)	2 (22.2)	3 (33.3)	2 (8.7)	9 (39.1)	5 (21.7)	5 (21.7)	2 (8.7)	0.143
I automatically pay more attention to patients who do not speak Dutch	5 (55.6)	3 (33.3)	-	1 (11.1)	-	9 (39.1)	9 (39.1)	2 (8.7)	3 (13.0)	-	0.391
I automatically spend more time on patients who do not speak Dutch	2 (22.2)	5 (55.6)	-	2 (22.2)	-	6 (26.1)	10 (43.5)	4 (17.4)	3 (13.0)	-	0.982

I consistently provide additional information to the patient	3 (33.3)	1 (11.1)	-	2 (22.2)	3 (33.3)	9 (39.1)	5 (21.7)	3 (13.0)	3 (13.0)	3 (13.0)	0.318
I think the issue of compliance is evident at my job	6 (66.7)	3 (33.3)	-	-	-	16 (69.6)	6 (26.1)	-	-	-	0.740
It is a habit of mine to keep my knowledge about compliance up to date by reading literature, visiting conferences, etc.	3 (33.3)	5 (55.6)	1 (11.1)	-	-	8 (34.8)	10 (43.5)	5 (21.7)	-	-	0.785
Additional questions											
Responsibility for compliance belongs to											
the orthoptist	1 (11.1)	4 (44.4)	-	2 (22.2)	2 (22.2)	3 (13.0)	13 (56.5)	2 (8.7)	5 (21.7)	-	0.292
the parent	7 (77.8)	2 (22.2)	-	-	-	15 (65.2)	6 (26.1)	1 (4.3)	1 (4.3)	-	0.439
the child	1 (11.1)	2 (22.2)	2 (22.2)	1 (11.1)	3 (33.3)	-	10 (43.5)	5 (21.7)	4 (17.4)	4 (17.4)	0.616
the government	-	-	3 (33.3)	1 (11.1)	5 (55.6)	2 (8.7)	5 (21.7)	6 (26.1)	5 (21.7)	5 (21.7)	0.054
the ophthalmologist	-	1 (11.1)	2 (22.2)	3 (33.3)	3 (33.3)	-	1 (4.3)	3 (13.0)	8 (34.8)	11 (47.8)	0.335
In the event of noncompliance, I put pressure on											
the parent	3 (33.3)	4 (44.4)	1 (11.1)	1 (11.1)	-	10 (43.5)	12 (52.5)	-	-	-	0.240
the child	1 (11.1)	4 (44.4)	1 (11.1)	-	3 (33.3)	3 (13.0)	13 (56.5)	4 (17.4)	-	2 (8.7)	0.303

Appendix VI) Results of the short compliance survey for all orthoptists in The Netherlands (Chapter 4)

The short compliance survey, which was sent to all Dutch orthoptists. It contained 37 questions and was subdivided into the following sections: "general information of the orthoptist," "concept of (non-) compliance," "attitude towards non compliance," "attitude towards non-native patients," "relationship between patient & orthoptist," and "managing non compliance." In total, all orthoptists of both groups, and 119 orthoptists from the NVvO completed the questionnaire.

	Number of orthoptists (151 in total) N (%)
General information about the orthoptist	
Work experience (years)	
<5	35 (23.2)
5 - <10	27 (17.9)
10 - <15	17 (11.3)
15 - <20	22 (14.6)
≥20	50 (33.1)
Workload per week (hours)	18:46 SD 7:10
I work at (multiple answers possible):	
a hospital in large city	40(26.5)
a hospital in a rural or suburban city	93 (61.6)
an academic hospital	14 (9.3)
other	13 (8.6)
Estimated % of non-Dutch speaking patients	22.45% SD 20.36
Perception of compliance and noncompliance	
Estimation of level of compliance in practice	73.75% SD 15.97
Noncompliance refers to *(multiple answers possible)*:	
not meeting the appointment	100 (66.7)
not making a new appointment	24 (16.0)
not complying the occlusion therapy	142 (94.7)
do not know	1 (0.7)
The patients in my practice often do not comply with treatment	
strongly agree	3 (2.0)
somewhat agree	5 (3.4)
neither agree or disagree	30 (20.1)
somewhat disagree	93 (62.4)
strongly disagree	16 (10.7)
do not know	4 (2.6)

Patients miss an appointment without notice fairly often	
(almost) always	0 (0.0)
quite often	5 (3.4)
sometimes	85 (57.0)
occasionally	48 (32.2)
(almost) never	11 (7.4)
Educational material can improve compliance	
(almost) always	25 (16.7)
quite often	64 (42.7)
sometimes	60 (40.0)
occasionally	0 (0.0)
(almost) never	0 (0.0)
do not know	2 (1.3)

	Number of Dutch orthoptists (151 in total) n (%)				
	Strongly agree	**Somewhat agree**	**Neither agree nor disagree**	**Somewhat disagree**	**Strongly disagree**
Attitude towards noncompliance					
I am able to recognize noncompliance	3 (2.0)	110 (73.8)	29 (19.5)	7 (4.7)	0 (0.0)
I find it difficult to talk with noncompliant parents about their attitudes	2 (1.4)	11 (7.4)	15 (10.1)	96 (64.9)	24 (16.2)
I find it difficult to talk with noncompliant children about their attitudes	4 (2.7)	3 (2.0)	11 (7.4)	92 (62.2)	38 (25.7)
It irritates me when parents do not comply with the prescribed treatment	2 (1.3)	33 (22.0)	60 (40.0)	47 (31.3)	8 (5.3)
When many patients do not show up for their appointments in one day, it influences my level of motivation at work	3 (2.0)	21 (14.3)	66 (44.9)	39 (26.5)	18 (12.2)
Attitude toward non-native patients					
Children who have parents not fluent in Dutch are less compliant than children of parents highly fluent in Dutch	2 (1.3)	43 (28.9)	61 (40.9)	39 (26.2)	4 (2.7)
I am tempted to provide further explanation to patients who do not speak fluent Dutch	22 (14.6)	74 (49.0)	49 (32.5)	6 (4.0)	0 (0.0)
Relationship between patient & orthoptist					
I have a good relationship with the parents of a child who receives treatment	11 (7.5)	130 (89.0)	5 (3.4)	0 (0.0)	0 (0.0)
I have a good relationship with the child	17 (11.6)	123 (83.7)	4 (2.7)	2 (1.4)	1 (0.7)
It is important that parents and children trust me	124 (84.4)	22 (15.0)	1 (0.7)	0 (0.0)	0 (0.0)
I provide the parents and child with reassurance that the amblyopia is being treated	84 (57.1)	62 (42.2)	1 (0.7)	0 (0.0)	0 (0.0)
	(Almost) always	**Quite often**	**Sometimes**	**Occasionally**	**(Almost) never**
Parents of children with amblyopia understand my explanation about amblyopia	65 (43.6)	79 (53.0)	5 (3.4)	0	0 (0.0)
Parents of children with amblyopia find its treatment important	49 (32.9)	96 (64.4)	3 (2.0)	1 (0.7)	0 (0.0)

	Strongly agree	Somewhat agree	Neither agree nor disagree	Somewhat disagree	Strongly disagree
Managing noncompliance					
Parents who ask many questions receive extra time and attention from me	31 (20.5)	93 (61.6)	27 (17.9)	0	0
I explain the consequences of noncompliance with occlusion therapy very well	97 (66.0)	47 (32.0)	3 (2.0)	0	0
I ask parents whether they understood my explanation	81 (55.1)	55 (37.4)	8 (5.4)	3 (2.0)	0
I ask whether occlusion therapy disrupts their daily lives	17 (11.6)	49 (33.3)	55 (37.4)	20 (13.6)	6 (4.1)
I explain the amblyopia treatment to the parents of the child	127 (86.4)	20 (13.6)	0 (0.0)	0 (0.0)	0 (0.0)
I explain the amblyopia treatment to the child	67 (45.6)	63 (42.9)	16 (10.9)	1 (0.7)	0 (0.0)
I direct my explanation specifically toward the child	1 (0.7)	27 (18.2)	56 (37.8)	56 (37.8)	8 (5.4)
I refer parents to the orthoptic website so they can read more information about amblyopia and its treatment	10 (6.7)	34 (22.8)	43 (28.9)	32 (21.5)	30 (20.1)

	Number of Dutch orthoptists (151 in total) n (%)
Managing noncompliance (continued)	
When verifying whether treatment had been carried out, I rely on information from (multiple answers possible)	
the parent	144 (96.0)
the child	125 (83.3)
increase in visual acuity	127 (84.7)
else	8 (5.3)
What measures do you take in cases of noncompliance with occlusion therapy? (multiple answers possible)	
I increase the occlusion hours	13 (8.6)
I decrease the occlusion hours	20 (13.3)
we have a serious conversation & make strict agreements	143 (95.3)
I switch over to atropine treatment	54 (36.0)
I plan the follow-up visit sooner than normal	107 (71.3)
else	53 (35.3)

Do you alert the child's general practitioner to noncompliance issues?	
yes, by telephone call	14 (9.3)
yes, with a letter	52 (34.7)
yes, else	-
no	84 (56.0)
What measures do you take if patients do not show up for their appointments? (multiple answers possible)	
I call them	31 (20.7)
the secretary call them	12 (8.0)
a letter will be sent	23 (15.3)
no action	101 (67.3)
Do you or your colleagues record which patients have not appeared for their appointments?	
yes	73 (48.7)
quite often	11 (7.3)
occasionally	20 (13.3)
no	46 (30.7)
Do you or your colleagues record whether a patient has come on referral from a child health care center after a positive vision screening test?	
yes	9 (6.0)
quite often	8 (2.0)
occasionally	6 (4.0)
no	132 (88.0)

Appendix VII) The Occlusion Patch Comfort Questionnaire (OCQ) (Chapter 5)

This questionnaire asks you for your feelings about four different brands of eye patches. It will be filled out for in total four times, the first three times during a phone call, the last time during the last house visit. The information you provide will be kept strictly confidential.

There are no right or wrong answers. Please choose the answer that is closest to your thoughts.

How long did your child wear this brand of eye patch?

Today: _____ (in hours or minutes)

Yesterday:_____ (in hours or minutes)

1) The patch my child wore was:	**Very large**	**Large**	**About right**	**Small**	**Very small**
2) The patch sticks to the skin of my child:	**Very strong**	**Strong**	**About right**	**Weak**	**Very weak**

	None	**Mild**	**Moderate**	**Severe**	**Very severe**
3) How much pain did your child have when removing the patch from the skin?	0	0	0	0	0
4) Wearing the patch makes my child's eye or eyelids red or irritated.	0	0	0	0	0
5) How red or painful was the skin of your child a few minutes after removing the patch?	0	0	0	0	0
6) How red or painful was the skin of your child more than a hour after removing the patch?	0	0	0	0	0
7) Has patching got a negative effect on the relationship between you and your child?	0	0	0	0	0
8) Was your child inconvenienced during patching?	0	0	0	0	0
9) How often is your child getting negative reactions from others when he/she is wearing the patch?	0	0	0	0	0
10) I notice that other children stare at my child when the patch is on.	0	0	0	0	0
11) I have trouble keeping the patch on my child.	0	0	0	0	0
12) My child gets upset because of patching.	0	0	0	0	0
13) I get upset because of patching.	0	0	0	0	0
14) Other family members get upset because of patching.	0	0	0	0	0

	Excellent	Good	Fair	Poor	Very Poor	Not Applicable
15) How well did the patch stick on the eye of your child?	0	0	0	0	0	
16) How well did the patch stick on the eye of your child during sportive activities (such as cycling, running, playing etc)?	0	0	0	0	0	0
17) How well did the patch stick on the eye of your child when it was warm or when he/she was sweating?	0	0	0	0	0	0
18) When your child also wears glasses: how well can the glasses be combined with the patch, without the ODM being attached?	0	0	0	0	0	0

	Strongly Agree	Agree	Neither Agree nor Disagree	Disagree	Strongly Disagree
19) My child does not seem to mind wearing the patch once it is on.	0	0	0	0	0
20) My child thinks the patch looks pretty.	0	0	0	0	0

21) How would you rate this brand on a scale from 0 till 10, where zero means very poor and ten means excellent, when looking at all aspects of this patch.

0 1 2 3 4 5 6 7 8 9 10

Appendix VIII) Summary of our questionnaire of Chapter 6

Summary of our questionnaire: the first ten domains were taken from the SPVA (The Netherlands Institute for Social Research 2007), the last domain contained questions taken from three other questionnaires about amblyopia.

Demography, Migration & Household: country of origin (parents and grandparents); age; gender; marital status; number of children; year and reason of immigration; immigrant integration course; number of people in household; quality of the residence.
Education: level of education (parents and grandparents); education abroad; education in the Netherlands; current education; kind of education.
Employment: current labor; year of first labor in the Netherlands; unemployment; switching jobs; making promotion; employee relation; entrepreneur.
Income: income; social support; amount of social support; financial worries; financial support for family abroad.
Health: current general physically and mentally health; general practitioner visits; medical specialist visits; hospitalization.
Language Usage: fluency in Dutch; speaking Dutch to partner and children; skills in speaking Dutch; skills in reading Dutch; skills in understanding Dutch people and Dutch television or radio; daily spoken language used.
Religion: kind of religion; frequency of practicing the religion; strict or lenient religious beliefs.
Family Bonds: contact with children who have left home; contact with other family members; giving help or advice to family members; close family bonds; care for grandparents; receiving care from grandparents; trust in and reliance on other family members.
Social Contacts: member of a club; contact with Dutch natives; contact with members from the same ethnic minority; having Dutch friends; neighbor contact.
Cultural Integration & Conceptualization: this domain contained distinctive Dutch opinions about: relationships between (Dutch) men and woman; responsibility of the man; responsibility of the woman; expectations of a son; expectations of a daughter; expectations of the relationships between elderly and younger people.
Lazy Eye: reason to patch; behavior of the child while patching; relationship between parent and child while patching; difficulties with patching; reactions of others about the patch; activities while patching; worries about amblyopia; worries about patching.

Appendix IX) The educational cartoon for occlusion therapy (Chapter 7 and 9)

Selected parts of the educational cartoon intended to improve compliance with occlusion therapy for amblyopia.

The cartoon consists of three parts which were bound together in one booklet (written permission for publication of fragments of this booklet was obtained).

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Part I: The cartoon story

As the cartoon focused mainly on the child, it was written from the perspective of a child aged between 3 and 6. Its purpose was to explain the principles of the lazy eye and its treatment, and the need for subsequent visits to the treating orthoptist. As most children treated for amblyopia are too young to read, it was designed without text. To enable each child to identify with the character in the cartoon, animal figures and other distracting visual elements were not included. The cartoons did not explicitly represent any ethnic or cultural group, and were depicted in black and white so children could colour them in according to their own interpretation. A few fragments of the story are depicted below:

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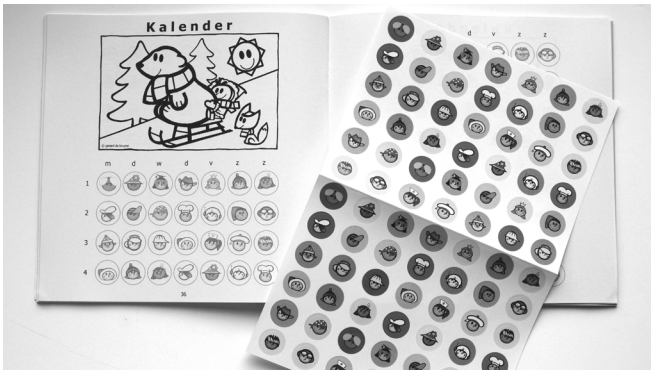
Part II: the calendar with stickers

The calendar and stickers served as a reward system: if the child was wearing the patch correctly that day, he or she was allowed to put a sticker on the calendar. The calendar covered a period of 84 weeks.



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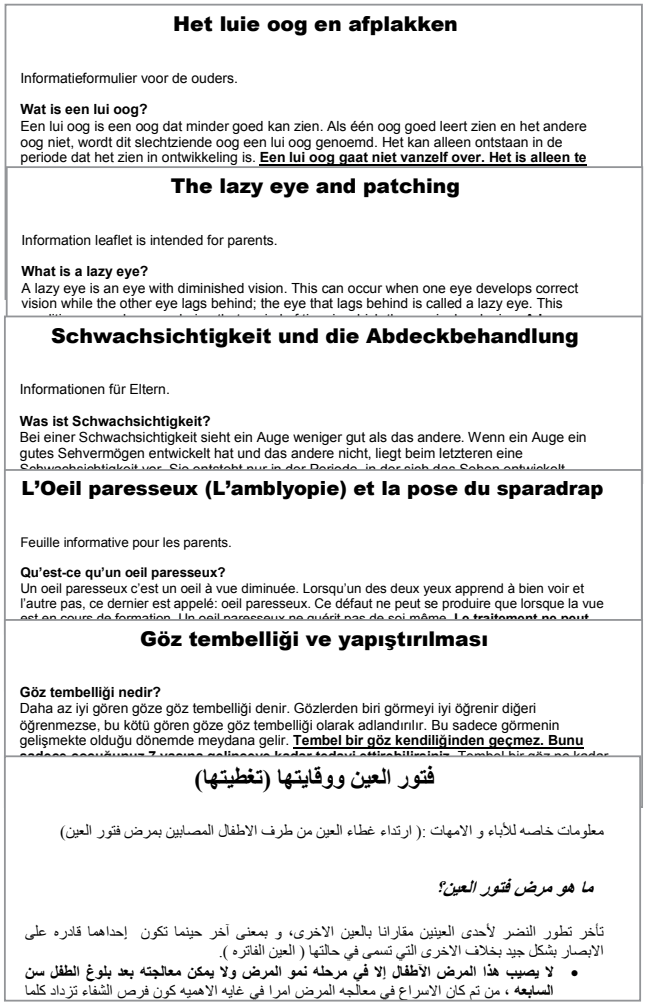
Part III: The parental information leaflet

As well as information about amblyopia and its causes, the leaflet describes the occlusion treatment and provides tips on patching. The information is available in six languages* (Dutch, English, French, German, Turkish and Arabic).



Het luie oog en afplakken

Informatieformulier voor de ouders.

Wat is een lui oog?
Een lui oog is een oog dat minder goed kan zien. Als één oog goed leert zien en het andere oog niet, wordt dit slechtziende oog een lui oog genoemd. Het kan alleen ontstaan in de periode dat het zien in ontwikkeling is. **Een lui oog gaat niet vanzelf over. Het is alleen te**

The lazy eye and patching

Information leaflet is intended for parents.

What is a lazy eye?
A lazy eye is an eye with diminished vision. This can occur when one eye develops correct vision while the other eye lags behind; the eye that lags behind is called a lazy eye. This

Schwachsichtigkeit und die Abdeckbehandlung

Informationen für Eltern.

Was ist Schwachsichtigkeit?
Bei einer Schwachsichtigkeit sieht ein Auge weniger gut als das andere. Wenn ein Auge ein gutes Sehvermögen entwickelt hat und das andere nicht, liegt beim letzteren eine

L'Oeil paresseux (L'amblyopie) et la pose du sparadrap

Feuille informative pour les parents.

Qu'est-ce qu'un oeil paresseux?
Un oeil paresseux c'est un oeil à vue diminuée. Lorsqu'un des deux yeux apprend à bien voir et l'autre pas, ce dernier est appelé: oeil paresseux. Ce défaut ne peut se produire que lorsque la vue

Göz tembelliği ve yapıştırılması

Göz tembelliği nedir?
Daha az iyi gören göze göz tembelliği denir. Gözlerden biri görmeyi iyi öğrenir diğeri öğrenmezse, bu kötü gören göze göz tembelliği olarak adlandırılır. Bu sadece görmenin gelişmekte olduğu dönemde meydana gelir. **Tembel bir göz kendiliğinden geçmez. Bunu**

فتور العين ووقايتها (تغطيتها)

معلومات خاصه للأباء و الامهات :(ارتداء غطاء العين من طرف الاطفال المصابين بمرض فتور العين)

ما هو مرض فتور العين؟

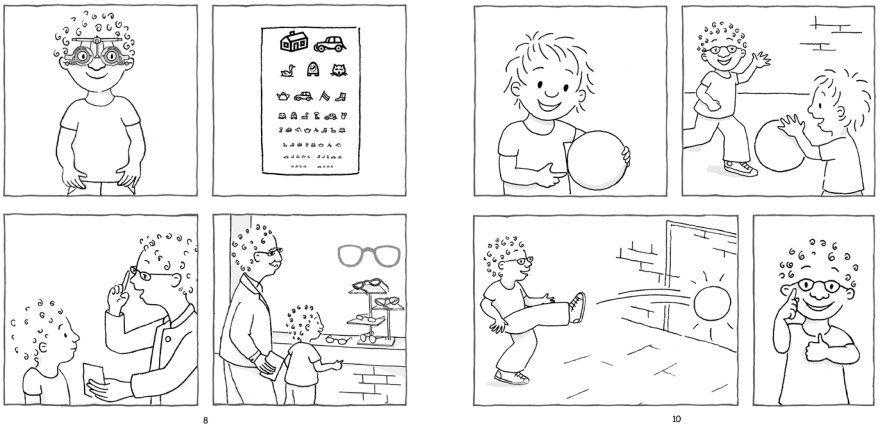
تأخر تطور النضر لأحدى العينين مقارانا بالعين الاخرى، و بمعنى آخر حينما تكون إحداهما قادره على الابصار بشكل جيد بخلاف الاخرى التي تسمى في حالتها (العين الفاتره).

- **لا يصيب هذا المرض الأطفال إلا في مرحله نمو المرض ولا يمكن معالجته بعد بلوغ الطفل سن السابعه** ، من تم كان الاسراع في معالجه المرض امرا في غايه الاهميه كون فرص الشفاء تزداد كلما

*) The authors acknowledge the contribution of dr. S.E. Loudon, Mr. P. Braakenburg, Mrs. B. Simonsz-Tóth, dr. E. Kilic and Mrs. Z. Al Attar for translating and/or editing the translated versions of the parental information sheet in English, French, German, Turkish and Arabic.

Appendix X) The educational cartoon for wearing glasses (Chapter 9)

Noncompliance with wearing glasses does occur. Therefore, an additional cartoon explaining to the child why he or she should wear glasses was created by the same artists that created the educational cartoon for occlusion therapy for amblyopia. The effectiveness of this cartoon was not demonstrated, but we did implement it together with the educational cartoon for occlusion therapy.



Appendix XI) Data collection and statistical analysis of Chapter 9

Observations in orthoptic practice

The recruited orthoptists in both groups were monitored by persons who did not participated in the treatment: in other words, during the observations, the researcher was not involved in the interactions between orthoptists and the patients. The orthoptists were not informed on which items they were being observed. Similarly, the observations were partly structured (i.e. the observer exactly recorded specific events). Structured observations were made with regard to the age and gender of the child, parents' fluency in Dutch, the time allotted per visit, the time actually spent per visit, and the time actually spent per explanation to the child and to the parent. Descriptive notes were taken on any explanation given regarding diagnosis and treatment to parent and child. Before implementation, each participating orthoptist was observed during an entire day, which resulted in approximately 15 patients that were observed per orthoptist. After implementation, orthoptists were observed for half a day.

Semi-structured interviews

The purpose of the face-to-face, semi-structured interview was to gain qualitative information about five different facets of the orthoptic practice: the organization of the clinic (such as the time allotted per new and per follow-up visit); collaboration within the clinic (e.g., between orthoptists and ophthalmologists or assistants); the demographic characteristics of their patients (e.g., the proportion of non-native patients); the orthoptists' attitude towards parents with a low SES or with poor fluency in Dutch; and the orthoptists' conception, attitude, and approach towards noncompliance. The interview was performed once before and once after implementation. Post-implementation, additional questions were asked on the educational cartoon, its use, and the course. Each interview lasted approximately one to one-and-a-half hour. This semi-structured interview was developed in a focus group (co-authors AMT, EV, MMS, WLA-T, and HJS).

Two researchers (WLA-T and AMT) separately analyzed all interviews according to the principles of Grounded Theory (Holloway 2005). All of the orthoptists' responses to each question were coded systematically and ordered according to whether the response represented a "*fact*," "*opinion*," or "*action*." This generated a systematic list of all possible statements per subject and how frequent they had been made by the orthoptists. The results of the analyses of both researchers were compared and discrepancies were discussed between the researchers and resolved.

Structured questionnaire

This questionnaire was developed in a focus group (co-authors AMT, MMS, and HJS), and consisted of two parts. The first part, "Orthoptists' individual personality features", contained questions to identify the personality of the orthoptist, and was subcategorized into "*autonomy*," "*being well organized*," "*ability to comprehend*," "*flexibility*," "*early adopter*," "*hierarchy*," and "*assertiveness*." This part of the questionnaire was based on categories that were selected from general assessments of healthcare givers' professional attitudes.

The second part of the questionnaire, "attitude towards noncompliance", consisted of statements that described different practical situations regarding noncompliance with occlusion therapy. These statements were developed on the basis of Prochaska & Velicer's Stages of Change model (Prochaska et al. 1997). The non-validated questionnaire contained 125 questions that were scored on a 5-point scale ("strongly agree" to "strongly disagree,") or a 6-point scale ("(almost) always" to "(almost) never,"). The structured questionnaire was applied twice before and twice after implementation.

Compliance survey

To assess a baseline and to detect national trends in the attitude towards noncompliance and in actions to detect and deal with noncompliance, a compliance survey was sent to all Dutch orthoptists annually for four years.

Approximately 95% of orthoptists in the Netherlands are members of the NVvO. Each year for four years, all NVvO-members (n = approx. 325) received annually a short, structured questionnaire survey developed in a focus group (co-authors AMT, WLA-T, MMS and HJS). It contained 37 questions, and was returned and processed anonymously, although later questionnaires returned by the same orthoptists could be recombined by reference to a self-generated code. The questionnaire collected information on the orthoptists: their conception of compliance and noncompliance; their estimates of noncompliance; the proportion of non-native patients in their practice; the actions they took to deal with noncompliance; their attitude towards noncompliance; and the relationship between patient and orthoptist. This survey was not validated.

Statistical analysis

All data was entered into SPSS 16.0 for Windows. *Chi-square* tests were used to evaluate differences in the baseline characteristics between the patients that were observed during the observations of the orthoptists in their practice pre- and post-implementation. Paired *T*-tests were used to compare the allotted time per visit, the time actually spent per visit,

and the time spent on explaining diagnosis and treatment to the child and parent both pre- and post-implementation. $P < 0.05$ indicated statistical significance.

Since the structured questionnaire was filled out multiple times over the years by the recruited orthoptists, we used multinomial multilevel models (*Proc Glimmix*) for ordinal questions to seek for differences in the answers given pre- and post-implementation. Questions having continues outcomes were analyzed with linear mixed models (*Proc Mixed*). Both models included a categorical variable indicating whether the questionnaire was filled out pre- or post-implementation, a variable indicating for each questionnaire the time elapsed since the start of the study, and the interaction between these two variables. We considered a *P*-value of 0.01 to be statistically significant to take multiple testing into account. All analyses regarding the structured questionnaire and survey were done using SAS 9.2.

Appendix XII) Detailed results of Chapter 9

Use of the educational cartoon

Twenty-three of all participating orthoptists had a favourable opinion of the cartoon and had received feedback from patients, most of whom were interested. Nine orthoptists (one working in a low-SES area, and eight working elsewhere in NL) believed that the cartoon had little effect, and had several negative opinions upon the cartoon. Table A12 shows the orthoptists' positive and negative opinions of the cartoon and their use of it. With regard to the distribution of the educational cartoon, all orthoptists said that explaining it required between two and five minutes more than they usually spent explaining the diagnosis and occlusion therapy. The time they spent distributing the cartoon also included the explanation about this study. Two orthoptists found that this took too much of their time; others did not find this disturbing, as they felt the cartoon made a valuable contribution to occlusion therapy (Table A12).

Orthoptists' awareness of noncompliance

Before implementation, orthoptists in low-SES areas estimated that their patients' compliance averaged 73.6%; after implementation, they estimated it at 54.8% (Fig. A12-1). Elsewhere in NL, orthoptists before implementation estimated their patients' compliance to be 75.6% and 66.8% after it.

After implementation, both groups of orthoptists characterized themselves as more aware of noncompliance than before implementation and more engaged in optimizing their patients' compliance (Fig. A12-2). During the implementation phase, they believed they were changing their routines to cope better with noncompliance than they had before it. However, these changes were not lasting: at the end of the study, orthoptists responded in much the same way as they had at the beginning.

Orthoptists' attitude regarding noncompliance

In the interviews and questionnaires completed before and after implementation, we were unable to demonstrate that the orthoptists' concepts and attitudes regarding noncompliance had changed. After implementation, they still had various ideas about the causes of noncompliance, and also a variety of perceptions of what compliance means.

Before implementation, they found noncompliant behaviour to be 'annoying', 'unpleasant' and 'hard to understand'. After it, they claimed to have a better understanding of their patients' noncompliant behaviour; overall, when noncompliance occurred, they pitied the child. One-third of the participating orthoptists – four in low-SES areas and five elsewhere in NL – believed that they were not responsible for the child's not being patched as prescribed, and believed that little actually helps to deal with noncompliance.

National compliance survey

In total, 285 orthoptists responded at least once to the national compliance survey, which was sent to all orthoptists in the Netherlands. One hundred seventy-two orthoptists returned the questionnaire in the first year, 157 in the second, 134 in the third, and 113 in the fourth. Overall, we were unable to detect any changes in their replies to questions on their concepts, awareness and attitudes regarding noncompliance. Neither did the actions orthoptists took to deal with noncompliance change over the years. Between 2006 and 2010, these 285 orthoptists estimated mean patient compliance with occlusion therapy to average 72% SD 13% each year (Fig. A12-1, P = 0.678).

Actions taken to deal with noncompliance

As reported previously in Chapter 4, during the semi-structured interviews before and implementation, each orthoptist referred to various methods of detecting and dealing with noncompliance. After implementation, they noted that they had shared these methods with one other during the interactive debates in the training course.

Actions taken to communicate with parent and child

Before implementation, orthoptists were observed in their practice for one day, accounting for 132 consultations with patients in low-SES areas and 344 consultations with patients elsewhere in NL. After implementation, we observed the orthoptists for half a day, accounting for 73 consultations with patients in low-SES areas and 195 consultations with patients elsewhere (Table 9.1, see Chapter 9).

When observing the orthoptists in their practices, we were particularly alert to any spontaneous suspicion of noncompliance in any of their patients. Both before and after implementation, we observed that only a quarter of orthoptists suspected noncompliance in a single patient during one day of observation; the others never did.

During these observations, the time taken for explanations to parents was the same after implementation as it had been before. Before implementation, orthoptists in low-SES areas spent an average of 2.5 minutes explaining diagnosis and/or treatment to parents; afterwards, they spent an average of 2.75 minutes (P = 0.832, Table 9.1). Elsewhere in NL, they spent 2.85 minutes on the explanation to the parent before implementation, and 3.1 minute after implementation (P = 0.796, Table 9.1).

The duration of the orthoptists' explanations to the child about treatment changed significantly after implementation. In low-SES areas, it was twice as long after implementation than before: 0'10" before vs. 0'24" afterwards (P = 0.061), against 0'26" vs. 0'40" elsewhere in NL (P = 0.121, Table 9.1). Observation of the orthoptists showed that, before implementation, none of the orthoptists in low-SES areas and only two elsewhere had directed their explanations about the treatment specifically to the child; afterwards, eleven more did so (P = 0.039): three in low-SES areas and eight elsewhere.

Before implementation, orthoptists from both groups asked parents 30% of the time whether their explanations had been understood; afterwards, they did so 60% of the time (P = 0.022, Table 9.1). However, the data obtained from the structured questionnaire showed no detectable change in communication styles towards parents.

Orthoptists in low-SES areas had more patients who did not speak Dutch well. To clarify their explanations, they used hardly any tools (such as pictures, posters, or medical interpreters); their explanations were often translated by a relative, sibling or friend who had accompanied the patient to the visit, or who had been telephoned during the visit. All orthoptists said that they did not use an official interpreter for patients who did not speak Dutch, as this was time-consuming and difficult to arrange. After implementation, orthoptists elsewhere in NL used drawings to support their explanation slightly more often (P = 0.022, Table 9.1).

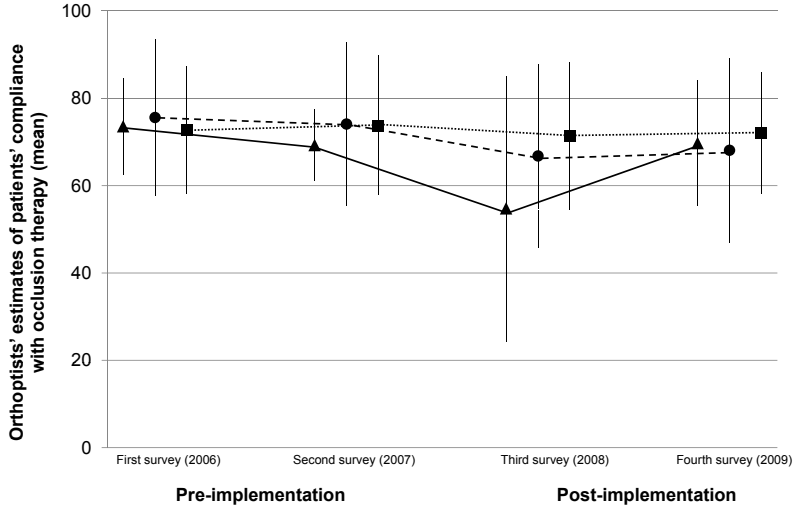


Figure A12.1) Orthoptists' estimations of patients' compliance derived from the national compliance survey that was sent annually for four years. We have reported earlier (Chapter 7) that electronically measured compliance with occlusion therapy was 52.0% before implementation and 62.3% after implementation.

Legends: Triangle (▲—▲) = mean percentages of patients' compliance with occlusion therapy as estimated by the participating orthoptists in low-SES areas. Round (●–––●) = as estimated by participating orthoptists elsewhere in NL. Square (⑤·····⑤) = as estimated by all Dutch orthoptists who were members of the Dutch Orthoptic association (*NVvO*), i.e. all remaining orthoptists and the orthoptists participating in the study.

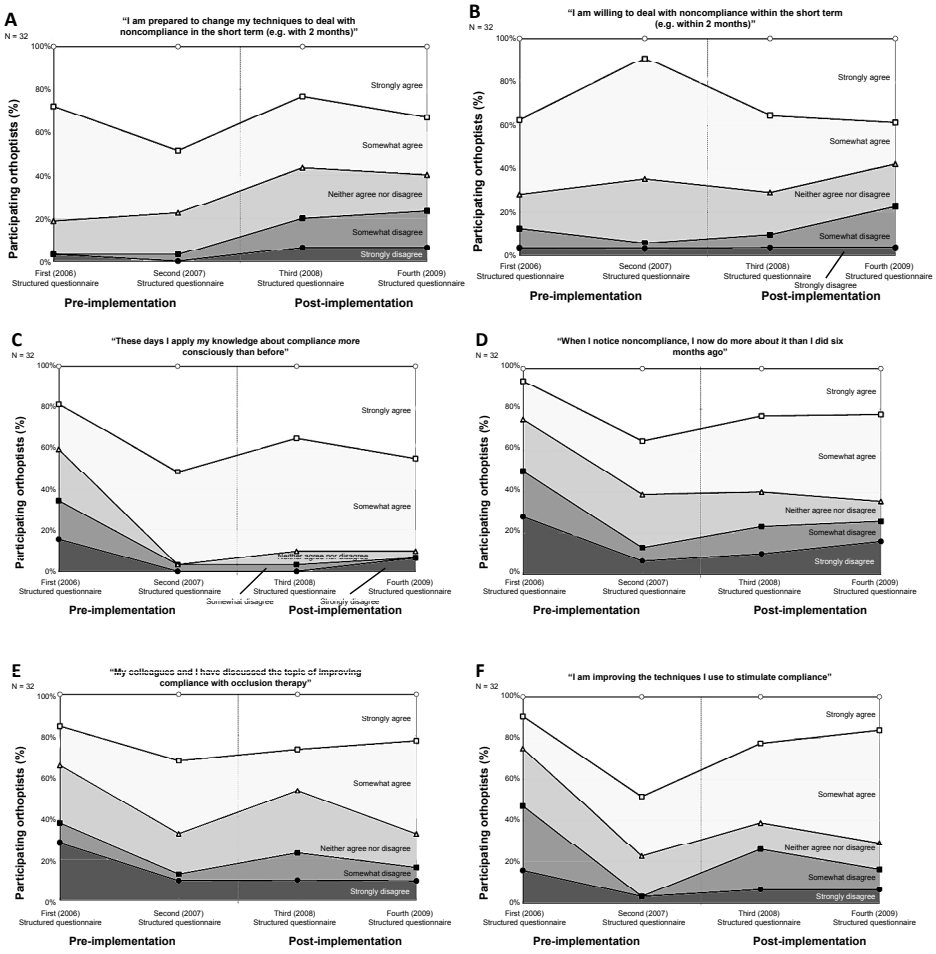


Figure A12.2) These graphs show the responses to questions in the structured questionnaire which was assessed four times during the study. Each graph shows the percentage of orthoptists (Y-axis) who strongly disagreed (dark-dark grey), disagreed somewhat (dark grey), neither agreed nor disagreed (grey), agreed somewhat (light grey) or strongly agreed (white) with the question over time (X-axis). The P-values indicate that responses had significantly changed throughout the time.

Table A12) Participating orthoptists' opinions of the cartoon and their use of it

	Low-SES areas (n = 9) N (%)	**Elsewhere in NL (n = 23)** N (%)
Negative opinions of the educational cartoon		
The cartoon is not attractive	0 (0.0)	2 (8.6)
I observe no clinical improvement with the use of the cartoon	1 (11.1)	4 (17.4)
Not all patients require the cartoon. It should therefore be given out only when there's a need for it.	0 (0.0)	1 (4.3)
Giving out the cartoon takes up a lot of my time	0 (0.0)	6 (26.1)
The cartoon will end up in the waste-paper basket	1 (11.1)	2 (8.6)
After a while, the children lose interest in the cartoon	0 (0.0)	8 (34.8)
Ortopad® eye-patch posters work better than the cartoon	0 (0.0)	3 (13.0)
Children are happier with a colouring page or balloon than with the cartoon	0 (0.0)	2 (8.6)
The cartoon is a typically girlish thing	0 (0.0)	4 (17.4)
Positive opinions of the educational cartoon		
The cartoon is attractive	5 (55.6)	9 (39.1)
I believe that the cartoon is of added value to patching treatment	5 (55.6)	7 (30.4)
The extra time needed to explain the cartoon when giving it out (max. 5 min) is acceptable	4 (44.4)	12 (52.2)
Giving out the cartoon takes up little of my time	2 (22.2)	0 (0.0)
Patients' responses to the cartoon are enthusiastic	3 (33.3)	10 (43.5)