9 General discussion

In the previous chapters several issues on various topics have already been discussed. The aim of this chapter is to bring some of these issues together to discuss them from a more general viewpoint and to introduce some new issues.

Health-related classifications

In this thesis, several names are used for the classification currently known as the International Classification of Functioning (ICF). Our studies were performed during the time that the changeover from ICIDH, via ICIDH-2, to ICF was ongoing, which may have led to some confusion. The ICF is one of the health-related classifications of the World Health Organisation (WHO) which was originally set up as the International Classification of Impairments, Disabilities and Handicaps (ICIDH) in 1980 [1] as a response to problems with evaluating health care at that time [2]. The ICIDH was developed because the even older International Classification of Diseases (ICD, first published in 1893) could not meet the need to describe medical outcome for chronic conditions. This ICIDH went beyond morbidity and mortality because it described the *consequences* of diseases and disorders. It recently underwent some revisions, which resulted in the ICIDH-2 in 2000 and the ICF in 2001 with an altered framework and terminology [3, 4]. General shortcomings of the ICIDH were insufficient attention to the role of the environment, overlap between dimensions and lack of clarity about causal and temporal relationships [2]. In the ICF positive counterparts of the ICIDH terminology are used; deviations or loss in body structures and functions are referred to as impaired body function and structure, difficulties with performing an activity are referred to as activity limitations (and not disabilities), and problems with participation or involvement in life situations are referred to as participation restrictions (and not handicaps). The current ICF terminology fits the ULAM better than the ICIDH terminology because the ULAM outcome measures are also positively expressed.

Health outcomes research

The changing view on health and health care has not only led to the development and revision of the WHO classifications, but also contributed to the rising of health outcome research. Within this discipline, it was increasingly acknowledged that methods and measures of activity (limitations) are very important indicators of human functioning and determinants of treatment effect [5-9]. Despite this, very limited attention is paid to devices like the ULAM and AM when it comes to reviews of methods and measures for health outcomes research. Scales and questionnaires are discussed extensively, whereas the technique of ambulatory monitoring is hardly ever mentioned although it comes up to a substantial number of requirements and criteria that are often formulated [9-11]. This may be due to the fact that ambulatory monitoring of activities is a relatively new technique that is still unknown to many and therefore not widely accepted. It may also be so that there is no notion of its numerous possibilities as yet because the technique goes beyond the general way of thinking about health outcome research. Hopefully, future technical developments and the increasing number of international publications involving ambulatory monitoring

devices such as the AM and ULAM will underline that ambulatory monitoring of activities is a relevant and valuable addition to the field of health outcomes research that can no longer be denied. It is important to realise, however, that this remark should not be confused with the idea that ambulatory monitoring is a new reference method and that measurement of what a person really does during everyday life is most important. As described several times in this thesis, the choice for an instrument should always depend on a complexity of factors.

Terminology for CRPSI

The results from the different application studies [12-15] showed that relatively large inter-individual differences with respect to the impact of upper limb CRPSI on everyday functioning exist. Such large inter-individual differences in presence, magnitude (and recovery) of limited upper limb activity as measured with the ULAM was also shown for impairments in upper limb CRPSI [13, 16, 17] and can be considered typical for disorders referred to as a syndrome. Regarding this heterogeneity in several aspects of the disorder, the 1994 decision of the International Association for the Study of Pain (IASP) to rename the 'disorder-with-an-ever-changing-name' to complex regional pain *syndrome* [18, 19] is considered appropriate. However, pain was not the most prominent impairment and neither did pain explain most of the variability in activity limitations in the studies described in this thesis. We therefore agree with vanderLaan et al [20], who suggested that it may be more appropriate to change the term pain to dysfunction, as in complex regional dysfunction syndrome type I (CRDSI). CRDSI is, in our opinion, also more appropriate with respect to the International Classification of Functioning (ICF) terminology because the consequences of this syndrome encompass all three levels of the ICF. The term dysfunction in CRDS can refer to consequences at all ICF levels whereas the term pain in CRPSI refers to only one of these levels.

Upper limb usage definition and framework

Our definition of upper limb usage as active movement(s) of (parts of) the upper limb(s) in relation to proximal parts, holding objects and/or leaning appeared to be workable when used together with the framework we compiled. Since developing and validating a novel device like the ULAM already is an innovative, and by that complex and time-consuming process, and because we did not know what the wearing comfort of the ULAM would be, particularly with sensors in the involved area, we wanted to limit the number and type of sensors in the first instance. In the framework used up till now, some sub-forms of upper limb usage were taken together because from a technical point of view, there are no real differences between for example leaning and holding, or between gesticulating, operating, handling and manipulating. As we also knew in advance that not all forms of upper limb usage could be equally well detected with the ULAM as a consequence of its technique, we made the assumption that a relationship exists between different forms of upper limb usage. Since leaning, holding and manipulating are usually preceded and followed by active upper limb movements to bring the limb in the right position to lean, hold or

manipulate in normal upper limb usage, a subject with an upper limb disorder will also perform less leaning, holding and manipulating with the involved side. Thus the assumption was that limitations of leaning, holding and manipulating are (indirectly) expressed in the amount of number of upper limb activity. This assumption and the technical-anatomical approach were, in our opinion, necessary for the developmental- and initial application-phase of the ULAM. Considering the results described in this thesis, sole usage of accelerometers can be sufficient to satisfactorily explore limitations of subjects with upper limb CRPSI. The ULAM outcome measures that have been used up to now were carefully formulated on the basis of those forms of upper limb usage from the framework that were best detected, as well as clinical considerations with respect to activity limitations in CRPSI. A more clinical definition of upper limb usage that may be worthwhile for further development and future applications, however, is active and purpose-directed usage of the upper limb (i.e. arm, including hand) to perform or carry out functional activities during everyday life. Using such a definition will result in a more extensive framework, which may also have consequences for ULAM requirements and the number and type of sensors and outcome measures. For example, if activity limitations of subjects who mainly have problems with manipulative upper limb usage (i.e. fine motor skills) have to be determined, the present ULAM outcome measures are too rough, and it may become necessary to distinguish between the sub-forms of primary functional upper limb usage gesticulating, positioning, handling and manipulating upper limb movements. Preliminary results with additional electromyography (EMG) sensors on the forearms to improve detection of these forms of upper limb usage have shown that there is some profit to gain [21, 22]. The development and validation of devices like the ULAM and its outcome measures clearly is an ongoing process of extending possibilities, optimising properties and enhancing interpretation.

Methodological considerations

Because the ULAM is, in fact, an extension of the AM, several limitations or issues that have already been discussed with respect to the AM [23] also apply to the ULAM. One of these issues is intra-subject variability of everyday activity, which refers to biological or natural differences in activity patterns of any given person between workdays, weekend days or irregular activities [24-26]. Between-day variability of upper limb activity may depend on the type of population that is studied; healthy subjects or subjects with a mild upper limb disorder may have a greater range of upper limb activity during everyday life than subjects with a severely limiting upper limb disorder. Second, between-day variability of upper limb activity may also depend on whether a disorder is acute or chronic. And third, between-day variability may differ depending on the outcome measures that are used. There may be differences regarding between-day variability between the intensity of upper limb activity and the percentage of upper limb activity or between the percentage of upper limb activity during sitting and the percentage of body motions that are performed (i.e. the ULAM outcome measure %dyn). From the viewpoint of novelty and patient burden (not

bathing, swimming etc) we chose to only perform 24-hour week-day measurements with the ULAM at first instance. However, it appeared that neither chronic nor acute CRPSI subjects had major problems with wearing the device for 24-hours. Retrospectively, it would have been interesting (or maybe even better) if we had performed some 48-hour measurements to have some indication of between-day variability of upper limb activity in CPRSI. Obtaining 'norm-values' for healthy upper limb activity and its between day between-day variability are also particularly important for future use as reference values to classify a subject with an upper limb disorder as limited or not.

Two other timely issues related to intra-subject variability of everyday activity are the impact of external factors and the ´reactivity-´ or ´perturbation-effect´. When performing a cross-sectional (comparison) study or longitudinal study with the ULAM, external factors possibly influencing activity patterns and upper limb activity, such as time of the year / season or family / living situation, should always be considered, because these factors may have an impact on validity of the results [27]. These factors are similar to the factor unrepresentativeness of the ULAM measurement period as already extensively discussed in chapter 8. This factor should also be taken into account during time management and planning of studies, selection of control groups and data-analysis. The second issue is the ´reactivity-´ or ´perturbation-effect´, which refers to the possibility that the ULAM influences the activity pattern of subjects because they are aware that they are measured [27]. Such an effect can also be due to wearing the instrument itself (i.e. a subject does not perform usual sports or therapeutic exercises while monitored). To avoid bias with this respect, instructions given to the subjects is of major importance as already explained in chapters 5 and 6. Another option to avoid some bias is to measure for more than one day and not use the data of the first day. Since it is questionable whether a subject is able to consciously adapt his or her activity pattern throughout the measurement, however, this option is probably unnecessary provided that it is always stressed that a subject's performance is not tested.

Last but not least, it has to be mentioned that ethical considerations of the ULAM are equal to those of the AM [23]. Just like all medical research, research the ULAM is subject to Medical Ethical Committee guidelines: the subjects can not be forced to participate and have to be well informed about the study including the consequences of participating. Measurement results may also not necessarily have consequences for their treatment, and it should be clear that measurements are not a test of a patient's capacity. It should be noted that the output of the ULAM is no more and no less than a specific set of outcome measures related to upper limb activity and mobility-related activities.

Practical aspects

Of the available methods for fixation of the acceleration sensors on the skin, Rolian Kushionflex™ or silicone-based stickers (Schwamedico) in combination with double-sided tape between these materials and the sensors appeared to be best, although this is still not optimal. To ensure that the sensors will remain fixed throughout the measurement period, mostly, additional skin friendly tape (Durapore™) was used on top the sensors and other fixatives. This was particularly important for the sensors on the forearm because sensors and cables at this location are more sensitive to bumping, getting wet and getting caught in objects. Moreover, since excessive sweating in the involved hand or arm may also be a problem in CRPSI, an extra roll of skin friendly tape was left behind for each subject if sensors and fixatives were in danger of getting loose. In two subjects that were studied in this thesis, a bandage was used for sensor fixation, however, because of allergy to all normally used fixatives and (rightly or wrongly) uneasiness about negative effects of taking off the fixatives. Because changes in sensor position during the measurement period are not desirable and should be kept to a minimum [23], subjects were instructed to check every now and then whether the sensors were still properly fixed. In case of (unexpected) problems of any kind, for instance with sensor attachment or fixation, ULAM power supply or recorder errors, the subjects were instructed to phone the researcher at any time during the measurement period, even in the middle of the night. Fortunately, this never happened.

Measurements with the ULAM may cause some discomfort because the system can not be used in a wet environment, which means that subjects can not shower or take a bath during the 24-hour measurement period. Cables or the recorder may disturb sleeping or (un)dressing, especially the cables of the ULAM forearm sensors that go up the sleeves and then down again to the recorder. For proper attachment of trunk and leg sensors with the AM, as well as with the ULAM, it was / is sometimes necessary to shave chest- or leg-hair, which did not cause difficulties. As for attachment of the ULAM forearm sensors, however, shaving may very well bring about objections. Fortunately, the majority of the subjects that have been studied so far was female, and the male subjects were either not very hairy or did not raise objections. Although some subjects disliked the ´tourist look´ and it sometimes appeared as though the weight of the recorder increased at the end of the measurement period, none of the subjects had insuperable problems with wearing the ULAM for 24-hours. Even though most subjects reported that they had to get used to wearing the device the first few minutes and again when they went to sleep, nobody found the ULAM uncomfortable to wear. So we do not think that wearing the ULAM influenced the subjects' activity. However, user-friendliness can and should always be subject to improvement. Especially if measurement periods for longer than 24 hours are desirable further technical developments such as a smaller, waterproof and lighter data logger, as well as (preferably wire-less) waterproof sensors are necessary.

Future applications and clinical implications

The work described in this thesis concerned the development, validation and application of the ULAM to determine activity limitations in subjects with upper limb CRPSI. The four application studies (chapters 5-8) have provided much information about the potential of this novel device. An important finding is that the consequences of upper limb CRPSI on everyday functioning are not restricted to impairments: clear limitations of upper limb activity were found. A clinical implication for medical practice therefore is the apparent importance to prevent upper limb CRPSI from becoming chronic because objectively measured limitations of the intensity, percentage and proportion of upper limb activity were found at on average 3.7 years after the causative event. It may be worthwhile to aim at reducing impaired AROM and strength in subjects with acute upper limb CRPSI because these impairments were the more important factors explaining variance in activity limitations in chronic CRPSI. However, this will have to be supported by further research. In a recent paper that described a positive effect of mono-disciplinary physical therapy to reduce impairments in acute upper limb CRPSI [17, 28-30], it was recommended that specific instruments suitable to measure activity limitations and participation problems need to be developed. In our opinion, the ULAM definitely meets this need. It has to be noticed, however, that only a subgroup within the CRPSI population was studied. Although the present population did not have severe uncommon motor impairments such as tremor, spasms or dystonia, one has to be very careful with generalising the present results to the total population with unilateral (or bilateral) upper limb CRPSI. Let alone to the total CRPSI population that consists for about 50% of lower limb CRPSI subjects. There may very well be discrepancies between other CRPSI subgroups or other upper limb disorders with respect to limitations of upper limb activity or the relationship between upper limb activity as measured with the ULAM and activity or impairment as measured with other instruments and their outcome measures. Such discrepancies and similarities may on the other hand also provide meaningful information for clinicians. Despite some limitations and some aspects that need further study, the ULAM has numerous possibilities for future applications in both the CRPSI population and other populations with upper limb disorders, such as repetitive strain injury, carpal tunnel syndrome, rheumatoid arthritis or after stroke.

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