

Introduction

Transverse mandibular and maxillary hypoplasia are relatively common entities seen in the Orthodontic and Oral and Maxillofacial practice¹. The origin of this hypoplasia lies in a disturbed ossification of cranial and facial bones during gestation and normal growth. Different factors that influence growth pattern include genetic, embryologic and trauma². The timing of ossification differs between mandible and maxilla. Via sutures the maxilla is attached to different craniofacial bony structures: nasal bone, zygomatic bones, lacrimal bones, inferior nasal conchas, palatine bone, vomer, frontal bones and ethmoids. The body of the maxilla consists of a left and right part connected at the intermaxillary suture. Rigid ossification is reached at around 15 years of age, although it has been suggested that complete ossification is to be reached after 30 years of age^{3, 4}. With regard to the mandible, fusion between the hemi-mandibles takes place at the mandibular symphysis, and complete ossification is reached at approximately 1 year of age³. The mandible is, unlike the maxilla, not directly attached to other bony structures, on both sides it is attached to the temporomandibular joint.

Developmental disorders and congenital craniofacial deformities are the main etiologic factors for transverse problems. Craniofacial deformities that are known to affect the transversal growth of the maxilla: cleft; frontal-nasal dysplasia, Pfeiffer's syndrome, Apert's syndrome and Saethre-Chotzen syndrome⁵. For the mandible, the following deformities can affect transversal growth: hypoglossia-hypodactyl syndrome, Nager syndrome and 18p syndrome^{6, 7}. The congenital deformity group consists of a rare, complex and heterogeneous group of patients for which extensive treatment plans are necessary which makes it difficult to compare outcomes. The focus of this thesis is mainly on non-congenital deformity patients.

Clinical symptoms that are associated with transversal hypoplasia of the jaws are generally related to either a discrepancy between a difference in tooth material and bone volume or a discrepancy between the upper and lower arches and may result in:

- Crowding, a condition in which there is inadequate arch circumference to accommodate teeth for proper alignment. Results of this misalignment are difficulties in maintaining adequate oral hygiene and aesthetical inconvenience.
- Crossbite, uni- or bilateral, a form of malocclusion where a tooth or teeth has a more buccal or lingual position than the antagonist tooth. As with crowding due to the position of teeth, oral hygiene may be compromised, and recession of the gingiva can occur.
- Buccal corridors, a black space in between teeth and the corner of the mouth that appears upon smiling. This can be esthetical disturbing to patients.

Correction of maxillary and/or mandibular transverse discrepancies in adolescent and adult patients gain increasingly more attention in the daily Orthodontic and Oral & maxillofacial practice. For small discrepancies tooth stripping or orthodontic treatment

alone are sufficient. Traditionally larger discrepancies (>4 mm.) are treated with tooth extractions and orthodontic treatment⁸. Advantages of this therapy are the relatively fast results and uncomplicated procedure. Disadvantages include the loss of healthy teeth, shortened dental arch and thinner lips¹. Ideally, extractions of intact teeth would be avoided. This can be done by widening the bony fundament e.g. mandible or maxilla followed by orthodontic treatment. This is what Angle et al. aimed for in the beginning of the 20th century, whereby in his vision an ideal occlusal scheme would prevent relapse of the orthodontic treatment¹.

Since the two segments of the mandible are fused at 1 year, stable non-surgical expansion is impossible⁹⁻¹¹. Due to the fact that ossification of the intermaxillary suture is much later in life, it is possible to expand the maxilla until approximately 15 years of age using orthodontic devices. This is known as Rapid Maxillary Expansion (RME). In the literature, good long-term results on RME have been reported^{12, 13}. In patients who are too old or where RME failed, purely orthodontic expansion of the maxilla is impossible. Then surgical techniques involving distraction osteogenesis are a possibility. For the mandible: Mandibular Midline Distraction (MMD); and for the maxilla: Surgically Assisted Rapid Maxillary Expansion (SARME) ^{14, 15}.

Distraction osteogenesis

In both SARME and MMD a method to induce both osteogenesis and histogenesis is involved: distraction osteogenesis. Initially introduced by Codivilla et al. at the beginning of the 20th century, however Ilizarov et al. refined and popularised the technique in lengthening lower limbs^{16, 17}. At the end of the 20th century, the technique was introduced in the maxillofacial region¹⁴.

In distraction osteogenesis, the normal fracture healing process is used to create new bone. Bone healing consists of four phases: I. inflammation phase; II. soft callus formation; III. hard callus formation; and IV remodelling¹⁸. Whereby the first phase ends around 7 days after the fracture occurred. A fibrovascular hematoma is formed and the collagen fibres are positioned parallel to the fracture, and the fracture is relatively unstable. During the soft callus phase, callus will be formed and replace the fibrovascular hematoma which adds to the stability of the fracture. The callus will be replaced by woven bone in the hard callus phase and during the remodelling phase woven bone will be replaced by lamellar bone and shaped to the original morphology.

The distraction osteogenesis technique involves an osteotomy, which initiates the fracture healing process. After the first healing phase a force is applied perpendicular to the osteotomy. This will gradually increase the gap between both sides of the osteotomy. A distractor may be used to apply this force. When the desired amount of lengthening or widening is achieved the distractor can serve as a stabiliser for the fracture until os-

sification is finished. In addition to the newly formed bone, soft tissue will also gradually expand due to tension on the tissues, and thus histogenesis will be initiated as well.

MMD

Upon the introduction of distraction osteogenesis in craniofacial region, expanding the mandible was challenging and unpredictable techniques were used. Orthodontist tried to widen the mandible with Schwartz appliances, lingual arches and functional appliances, however, long-term results show high relapse rates¹⁹. Surgeons have tried an alternative approach where a vertical osteotomy was made and the hemi-mandibles were positioned laterally with or without a bone graft^{19, 20}. Due to the risk of periodontal problems, lack of rigid fixation, the requirement for a bone graft and risk of relapse this technique was rarely indicated. In 1948 Crawford presented a rudimentary distraction technique, whereby after a mandibular symphyseal fracture with gradual traction mandibular widening was achieved²¹. Guerrero et al. were the first to present a technique with distraction osteogenesis¹⁵.

In general, MMD is performed in a hospital setting, where the surgery itself is performed under local or general anaesthesia, usually the latter^{22, 23}. Local anaesthesia is infiltrated in the buccal fold. A horizontal or vertical incision is made in the mucosa at the buccal fold to get access to the mandible. The mucoperiosteum is reflected and an osteotomy is performed using a reciprocating saw and finished with an osteotome in the region of the dental roots. In general, a midline split is performed, however, paramedian or stepwise osteotomies have been described. A tooth-borne distractor has been placed by the orthodontist prior to the surgery or a bone-borne is placed during surgery. After a latency period of 5-7 days, the distractor is activated with a rate of 0,5-1mm/day, until the desired amount is achieved. A consolidation period is ideally held for 2-4 months after which orthodontic treatment is initiated, however immediate start shortly after distraction has been described^{24, 25}. An alternative approach is the floating bone concept, hereby the teeth next to the distraction are not attached to fixed appliances and move or 'float' into the distraction gap.

Although it has been reported that MMD as a treatment modality can effectively widen the mandible, most of the follow-up studies are limited to the first year and/or were presented as case series²⁶⁻²⁸. The long-term stability is an essential part of the discussion to either choose for extraction therapy or widening. Therefore, it is viable to gain more insight in the long-term dental and skeletal effects of MMD.

Distractors can be applied on teeth, bone or a combination (hybrid). A bone-borne distractor is directly attached to the bone on either side of the osteotomy. A tooth-borne distractor is applied to teeth, usually a premolar and molar on each side. Both distractors have their advantages and disadvantages. With bone-borne distractors a second surgery is necessary for the removal of a bone-borne distractor, causing inconvenience

for a patient and more costs. A tooth-borne distractor applies the distraction force directly to the teeth, this might induce dental tipping and movement out of the alveolar bone, which could attribute to more relapse and periodontal problems. The hybrid distractor shares both the dis- and advantages of the bone- and tooth-borne distractor, in addition, the biomechanical effects are in between the bone- and tooth-borne distractors²⁹.

Due to the variability of the designs, the skeletal effects may vary as well. Mainly the vector and rigidity of a distractor attribute to the skeletal effects²⁹. Skeletal tipping is the effect whereby cranially more widening is obtained than caudal. It is thought that a less rigid distractor allows for more skeletal tipping, as the hemi-mandibles have more freedom to move³⁰. In addition, a tooth-borne distractor applies its forces above the centre of resistance causing more skeletal tipping. A result of skeletal tipping is that less basal bone is expected to form, which could increase the risk for relapse¹⁹.

In MMD the distractor is on both sides attached to a hemi-mandible which is connected to the temporomandibular joint (TMJ). Although the TMJ is well encapsulated and positioned by the masticatory muscular complex, movement in the joints is expected as a result of the distractor vector. As this would increase stress on the joints, they will have to adapt³¹. A fine element study presented by Kim et al. showed a tendency of higher stress levels at the articular disc for tooth-borne devices than for the bone-borne devices.²⁹ Therefore, a rigid bone-borne distractor was designed the 'Rotterdam Mandibular Distractor' which would theoretically reduce skeletal tipping and stress on the TMJ. This assumption will be assessed in this thesis.

SARME

The first reports of expanding the maxilla date back to the 19th century, where thumb pressure and the use of a C-spring were described³². Angell et al. were the first to describe the use of a jackscrew to expand the maxilla, a basic form of rapid maxillary expansion (RME)³².

In literature, the first to describe a surgical technique to expand the maxilla was Brown et al. who performed a midline split. Initially, it was thought that the intermaxillary suture was the centre of resistance³². However, during the 1970s it was demonstrated that the zygomaticofrontal, zygomaticotemporal and zygomaticomaxillary sutures were the actual center of resistance³². This resulted in adjustments of the osteotomy sites. The following osteotomies were added to the midpalatal osteotomy: piriform aperture, zygomatic buttress and pterygoid junction. As this technique gained more attention different variations appeared and different authors advocate different techniques.

In the 1980s the first reports describe the use of a hyrax in combination with osteotomies to expand the maxilla using distraction osteogenesis, surgically assisted rapid maxillary expansion³³. In the 1990s various bone-borne appliances were introduced to expand the maxilla. The bone-borne distractors were introduced as it was hypothesised

that tooth-borne appliances would cause more dental and skeletal tipping, resulting in more periodontal problems and relapse. However, Koudstaal et al. showed that there were no biomechanical differences between the bone- and the tooth-borne distractors.³⁴ In addition, a stable widening was achieved after the first 12 months for both groups. However, little is known on the long-term effects of SARME, the long-term stability is an essential factor in deciding between SARME or extraction therapy.

Nowadays the procedure is generally performed under general anaesthesia, although some authors mention performing the procedure using local anaesthesia. Either a tooth-borne hyrax appliance is applied pre-operatively by the orthodontist or a bone-borne distractor is placed during surgery. First, the mucosa will be infiltrated with a local anaesthesia with a vasoconstrictor. Then an incision is made in the buccal fold, just 3-4mm above the muco-gingival boundary from the first premolar to the first premolar. The mucosa is reflected and a clear view on the maxilla and access to the nasal aperture is obtained. An osteotomy is made on the level of LeFort I and in the midline, this can be done with the use of a combination of the following instruments: burr, reciprocal saw or osteotome. During surgery, the surgeon will pay attention not to damage the roots of the teeth. Mobility of the two maxillary parts is verified and after rinsing the wound it is closed with resorbable sutures. In addition to the above mentioned surgical steps, some authors advocate the release of the nasal septum and the pterygomaxillary junction.

Following a latency period of 5-7 days, the distractor will be activated until the desired amount of widening has been reached. A consolidation phase of 3 months is generally kept before the distractor is removed. In general, after a period of 2-4 months the orthodontic treatment is initiated.

As the emphasis should lie on minimising morbidity, a new technique emerged which uses miniscrew-assisted rapid palatal expansion³⁵. This technique uses a hybrid-distractor which is applied on teeth and on two or four mini-screws placed paramedian in the hard palatal bone. The main benefit is that no osteotomies are made, reducing the risks of the surgical intervention. This technique is an alternative for young adult patients who need expansion of maxilla. This is shown in a group of patients with an age ranged from 19 and 26 whereby the distractor forces widened the intermaxillary suture in 86.8% of the patients³⁶. The first reports are promising, however the technique will not be part of this thesis.

Complications

SARME and MMD both consist of a combined orthodontic treatment and surgical intervention where the patient sees both the orthodontist and the oral and maxillofacial surgeon for at least 1 year. It is necessary for patients to realise what they can expect during this period, for them to make a balanced decision. Important factors to consider other than technical effectiveness, are complication rates and patient experiences.

The amount and burden of complications in SARME are well described^{32, 37}. In general, these complications are regarded to be mild and include haemorrhage, pain, loss of tooth vitality and damage to the infra-orbital nerve. Regarding MMD, complications were reported aside of the main biomechanical effects and therefore the usability is minimal³⁸. In addition, the studies specifically aimed at complications did not describe the complications systematically³⁹. Systematically analysing surgical complications is important to enable comparison of clinical outcome and complications from different research groups.

Patient experience

Although both SARME and MMD are relatively commonly performed procedures, little is reported on patient expectations, experiences and satisfaction^{40, 41}. Aside from the orthodontic treatment, it is expected that in the post-operation period patients can experience swelling and pain. During the distraction and consolidation phase, a possibly aesthetically disturbing diastema between the upper or lower incisors appears and the distractor device might interfere with speech and cause pain and pressure ulcers. It is obvious that this is uncomfortable for a patient. It is, however, unknown how patients perceive this discomfort. As this information is essential to properly inform patients before surgery, this will be studied in this thesis.

The effects of orthognathic surgery on the appearance of the face, the soft tissue effects, gain increasingly more attention. As in dental implant surgery a widespread saying is 'The bone sets the tone, the soft tissue is the issue' this is also the case in orthognathic surgery, only in another sense. As the bony structures more or less dictate the position of the surrounding soft tissues. Therefore, interference with facial bony structures can affect to some extent facial appearance. In both SARME and MMD new bone is created in either the maxilla or mandible and thus facial changes are expected. With regards to SARME Nada et al. showed that a posterior positioning of the upper lip, increased cheek projection and increased nose volume are seen after distraction⁴². In addition, Xi et al. showed that a posterior and inferior movement of the maxilla results in an auto-rotational movement of the mandible⁴³. As a consequence of this auto-rotation, a more pronounced chin is expected.

With regards to the soft-tissue effects of MMD, little is known. Bianchi et al. did a soft tissue analysis in patients who underwent both SARME and MMD⁴⁴. They concluded that the combined effects of both treatment modalities were aesthetically satisfactory as buccal corridors were reduced and a 'pleasant' fullness of the mouth was observed. The methods of the study presented by Bianchi et al. lack a proper description of how this was measured and objectified⁴². In addition, the data section showed a transverse increase in the cheek, mouth and chin region. However, as Xi et al. reported a more pronounced chin in patients who underwent SARME, the results regarding the chin region

might be biased due to SARME procedure. Although soft tissue analysis is not a part of this thesis, patient experience and satisfaction are related to each other and will be studied in this thesis.

GENERAL AIM AND OUTLINE OF THE THESIS

The general aim of this thesis is to assess the long-term dental and skeletal effects of MMD and SARME and will be outlined in chapter 5 and 6. Koudstaal et al. and Verstraaten et al. presented comprehensive reviews on SARME^{32, 45}. In addition to these reviews, in chapters 2 and 3 systematic literature reviews are provided regarding MMD in general and another regarding the effect of MMD related to three-dimensional imaging techniques. Although MMD is considered as a safe and effective treatment modality, relapse, distractor type and patient experience are poorly studied topics. In addition to the general aim of this thesis, the following topics will be addressed:

1. In chapter 4 the hypothesis that a rigid bone-borne distractor would reduce skeletal tipping and stress on the TMJ's is studied. A new rigid bone-borne distractor introduced and compared with a non-rigid distractor.
2. Only few studies have been performed to assess the complication rate of MMD. In chapter 7 the Clavien-Dindo complication classification system is used to systematically assess the complications in MMD using a bone-borne distractor.
3. Complimentary to this study, a study assessing patient experience and satisfaction during and after SARME and MMD is presented in chapter 8.
4. The initial performed studies used conventional radiographs, however, nowadays 3D imaging techniques enables better interpretation of the skeletal and soft tissue changes after SARME and/or MMD. A systematic review is performed to assess what has been studied regarding MMD using 3D imaging techniques will be outlined in chapter 3.

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