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Is early osteodistraction a solution for the ascending ramus compartment in hemifacial microsomia? A literature study

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SUMMARY. Aim: **In hemifacial microsomia, osteodistraction before skeletal maturation claims to induce both bone- and soft-tissue generation in such a way that conventional bone grafting and soft-tissue grafts are not necessary. Early osteodistraction in facial microsomia would have a positive effect on the 'functional matrix', and allow symmetrical vertical and sagittal expansion of the midface and mandible. The aim of this literature survey was to find evidence for this hypothesis by analysing long-term follow-up reports on distraction histiogenesis in the ascending ramus.** Material: **Only eight published studies were found, of which only two had more than ten patients, two were case reports, and three were from the same institution.** Result: **Invariably, the results pointed towards over-correction, repeated osteodistraction procedures, soft-tissue stretching (but no lateral augmentation), and to soft-tissue complications. The studies did not allow a conclusion to be made as whether increased vertical gain in the ascending ramus was unstable because of decreased growth on the affected side, inborn or iatrogenic, or due to resorption of the bone generated by distraction. To date, there is no evidence that osteodistraction produces better results and has lower morbidity than conventional growth centre transplantation and separate soft-tissue augmentation.** Conclusion: **Recommendations for prospective studies are: sharp differentiation between the four Pruzansky–Kaban mandibular types, multi-centre study of a surgical protocol to increase the sample number using a standard three-dimensional evaluation protocol, and differentiation between decreased growth and collapse of the newly generated bone.**

INTRODUCTION

Hemifacial or facial microsomia is the second most common facial birth disorder after cleft lip and palate (*Murray* et al., 1985), with an incidence of 1/3500–6000 live births (*Grabb*, 1965; *Poswillo*, 1974b). The condition is bilateral in 10% of cases (*Converse* et al., 1977). Reconstruction of the malformed hard and soft tissues of the microsomic face is a difficult task which is usually staged over many years. The choice of an appropriate treatment concept has therefore always been of concern to craniomaxillofacial centres. The tissue deficiency in the ascending ramus (postero-lateral face), composed of the ascending mandibular ramus, the muscles of mastication and the integument, remains especially challenging.

From the mid-1970s till the mid-1990s, costochondral grafting (*Poswillo*, 1974a; *Samman*, 1996; *Padwa* et al., 1998) was a widely adopted technique to reconstruct a deficient ascending ramus in Pruzansky–Kaban types IIB and III pre-pubertally (Table 1; *Pruzansky*, 1969; *Mulliken* and *Kaban*, 1987; *Kaban* et al., 1988). Less-affected children (mandibular types I and IIA) were not treated until adolescence (*Obwegeser*, 1970, 1974), or received 'functional' orthodontic appliances and early mandibular osteotomies to keep up with vertical midfacial development (*Kaban*, 1990). After skeletal correction, the soft-tissue deficiency in the posterior face was initially corrected with fat or derma-fat transplantations (*Davis*, 1968) and later with free microvascular flaps (*LaRossa* et al., 1980; *Kaban*, 1990, *Siebert* et al., 1996).

Since 1995, distraction osteogenesis has been applied increasingly in young patients to induce bone- and soft-tissue generation. Both extra-oral (*McCarthy* et al., 1992, 1997; *Molina* and *Ortiz Monasterio*, 1995; *Kaban* et al., 1998) and intra-oral (*McCarthy* et al., 1995, 2001; *Diner* et al., 1996 and 1999) techniques have been used. It was proclaimed that distraction osteogenesis would affect the entire facial milieu with an increase in soft-tissue envelope due to expansion and muscle hypertrophy (*McCarthy*, 1994; *Molina* and *Ortiz Monasterio*, 1995; *Rachmiel* et al., 1995; *McCarthy* et al., 1998). *Molina* and *Ortiz Monasterio* (1995) stated that 'The skeletal distraction proceeds parallel to an expansion of all the soft tissues of the face and upper neck (skin, muscles, vessels and nerves), achieving aesthetic results much superior to those obtained by skeletal surgery, by soft-tissue surgery done independently, or by a combination of both'. The photographs in their article are indeed convincing. However, recent follow-up studies of early distraction before skeletal maturation (before the permanent dentition is

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Table 1 – Pruzansky–Kaban classification of the mandibular deformity in hemifacial microsomia (*Pruzansky*, 1969; *Kaban* et al., 1988; *Oeltjen* et al., 2001)

Type I	All mandibular and TMJ components are present and normal in shape but hypoplastic to a variable degree as compared with the contralateral side
Type IIA	The mandibular ramus, condyle, and TMJ are present but hypoplastic and abnormal in shape. The mouth can be symmetrically opened
Type IIB	The mandibular ramus is hypoplastic and markedly abnormal in form and location, being medial, anterior and inferior. There is no articulation with the temporal bone
Type III	The mandibular ramus, condyle, and TMJ are absent. The lateral pterygoid muscle and temporalis, if present, are not attached to the mandibular remnant

TMJ: temporo-mandibular joint.

Table 2 – Management guidelines for early mandibular osteodistraction in hemifacial microsomia (*McCarthy* et al., 2001)

Age	Indication according to mandibular Pruzansky type/symptoms
0–2 years	Severe airway obstruction (to prevent tracheostomy)
2–6years	Severe types I and II; associated sleep apnoea; distraction of the costochondral graft in type III
6 years–adolescence	Sleep apnoea; patient had never received mandibular reconstruction; distraction of the costochondral graft

complete) have shown variable limitations in the skeletal correction and recurrence of the original skeletal deformity. Soft-tissue complications and relapse of the soft-tissue augmentation also result. The aim of this study was to critically appraise these follow-up reports on the long-term effectiveness of early distraction osteogenesis techniques in the correction of the ‘ascending ramus compartment’ in facial microsomia patients. All data presented are related to unilateral distraction.

LONG-TERM STABILITY OF THE DISTRACTED ASCENDING RAMUS

Four cases of mandibular corpus distraction were reported in the first half of the 20th century (*Rosenthal*, 1930; *Wassmund*, 1935; *Kazanjian*, 1941; *Crawford*, 1948). The technique only received attention at the end of the century with *McCarthy's* (1994) report on a small series of early mandibular osteodistraction in hemifacial microsomia cases. The technique became an accepted method of treatment worldwide with the advent of bi-directional distraction (*Klein* and *Howaldt* 1995, 1996), enabling the surgeon to expand both the horizontal and vertical rami.

Vertical ramus distraction in hemifacial microsomia when performed early is increasingly reported in the literature (*McCarthy*, 1992; *Molina* and *Ortiz-Monasterio*, 1995; *Polley* and *Figueroa*, 1997; *Tharanon* and *Sinn*, 1999; *Appel* et al., 2000; *Smith* and *Harnish*, 2001). In 2001, *McCarthy* et al. proposed a protocol for osteodistraction in infants (Table 2). Evidence of the efficacy of early distraction from long-term follow-up reports is relatively rare. A total of eight reports were found at the time of writing this manuscript, three of which were submitted by a single institution, with probable overlap in patient inclusions. Only two reports included more than ten patients, and two were case reports. The heterogeneity of the condition, and the relatively low incidence are partially responsible for the problems with these studies.

Molina and *Ortiz Monasterio* (1995) published their results on 65 patients with hemifacial microsomia treated by early extra-oral distraction. Patients were grouped according to the Pruzansky–Kaban classification (groups I, IIA and IIB). Twelve patients older than 14 years were included in the study and it was not indicated to which groups they belonged. The average follow-up period was 19 months (range, 3 months – $3\frac{1}{2}$ years). Anthropometrical measurements were used for the assessment. The authors tabulated the post-distraction measurements, but failed to report the late follow-up measurements. They stated that ‘in the younger group, facial development on the unaffected side proceeds normally, growing more than the distracted side’. Therefore, the authors propose over-correction and second-stage distraction. *Grayson* et al. (1997) studied the results they obtained in five patients, followed for a period of 1–6 years. The age at time of surgery is not mentioned as such in the article, but it concerned early distraction, as defined above. Whilst the measurements were taken from serial posterior–anterior (p.a.) and lateral cephalograms, other data (rate or ratios) were not supplied. Regarding the vertical ramus, they stated that ‘the increase in height over 5 years of growth is greater on the side that was not distracted, over-correction is therefore recommended in the growing child’. *Hollier* et al. (1999b) followed eight patients, operated on at age of $1\frac{1}{2}$–$3\frac{1}{2}$years, with serial p.a. cephalometry to evaluate the change in vertical ramus height. Clinical follow-up averaged 32.6 months (range 12–92 months). The results were reflected in ratios. Following distraction, growth of the affected vertical ramus showed a slower rate than that of the unaffected side in each case. Therefore, the authors readily accept that ‘to minimize the need for repeat distraction, it would seem intuitive that over-correction would be beneficial’.

The amount of relapse could not be correlated to the amount of distraction, degree of asymmetry, follow-up time, or age, because the small numbers did not allow statistical analysis. *Kusnoto* et al. (1999), studied growth parameters of six patients with an average age of 12.5 years (SD 2.4 years) at the time of distraction. They were followed for 8 years preoperatively and $1\frac{1}{2}$ years post-distraction, with p.a. and lateral cephalograms. Most of the favourable growth effects were found in the body of the mandible rather than in the ramus. On average, the distracted ramus shortened by 1 mm during follow-up. *Marquez* et al. (2000) used a.p. lateral cephalograms, and panorex views, to measure the 2-year post-distraction changes in the posterior face of a 7-year-old boy. From the pictures, one can deduct that it concerned a type IIB mandible. There was a relapse of 87% of the 15 mm of vertical distraction, though the a.p. distraction gain was stable. They concluded that 'distraction osteogenesis does not accelerate growth and does not predictably increase length in the mandibular ramus'. *Appel* et al. (2000) treated three patients of 10-, 11- and 12-year of age with osteodistraction and followed them for 22, 16 and 10 months respectively. They found no relapse after 24, 32 and 25 mm of distraction. The article does not mention the severity of the deformity nor the methods of investigation. *Stucki-McCormick* et al. (2001) reported long-term follow-up of one case operated on early, without giving specific ages. The patient probably had a IIB-type mandible. She concluded that the bony improvements which resulted were not maintained during growth. *Rachmiel* et al. (2001) also noted 'some relapse' in vertical ramus height in 11 patients after 1 year (Pruzansky–Kaban types I and IIA), when operated on at an age of 6–12 years. The results were obtained from p.a. cephalograms and reported as ratios.

Investigators tend to mix the terms 'rate' and 'ratio', which makes comparisons difficult. (When both sides of the mandible grow at the same rate, the ratio will become smaller with time. When the ratio remains constant, this means that the affected side grows at a slower rate.)

LONG-TERM RESULTS OF THE SOFT-TISSUE AUGMENTATION BY BONE DISTRACTION IN THE POSTERIOR FACE

Before the advent of osteodistraction, soft-tissue augmentation in the posterior face was usually done after skeletal reconstruction (*Murray* et al., 1985; *Posnick*, 1998). Some of the pioneers of osteodistraction for hemifacial microsomia still claim that the surrounding soft tissues (skin, muscles) also grow due to lengthening (soft-tissue expansion) and increased volume (muscle hypertrophy; *Takato* et al., 1993; *Molina* and *Ortiz Monasterio*, 1995, 1997; *Pensler* et al., 1995; *Diner* et al., 1996; *Polley* et al., 1997; *McCarthy* et al., 2001). If this was so, secondary soft-tissue augmentation would be less necessary, at least in mild cases. The issue remains controversial. *Marquez* et al. (2000), in their single-case follow-up report, stated that the soft-tissue matrix decreased in response to the skeletal enlargement by osteodistraction. The mechanism is similar to that described after conventional surgical correction, namely that due to the midline move to the contralateral side, the soft-tissue contour on the non-affected side seems to increase, while the already hypoplastic soft tissue on the affected side is further stretched and attenuated (*Kaban* et al., 1998). Some still use free dermis-fat grafts for soft-tissue augmentation on the affected side (*Hollier* et al., 1999a). There is also a report of simultaneous correction of bone and soft tissues with osteodistraction and a parascapular osteocutaneous flap in a Pruzansky Grade III case (*Polley* et al., 1996).

Some soft-tissue complications following distraction are well known and with extra-oral appliances, pin tract infections and hypertrophic scars have been reported (*Rachmiel* et al., 1995; *Klein* and *Howaldt*, 1996; *Corcoran* et al., 1997; *Carls* and *Sailer*, 1998; *Tharanon* and *Sinn*, 1999; *Hollier* et al., 1999a). With buried intra-oral appliances, soft-tissue infections are also known to occur as soft-tissue closure is difficult (*Diner* et al., 1999).

DISCUSSION

Most of contemporary literature on facial microsomia favours mandibular reconstruction before skeletal maturation (*Munro* and *Lauritzen*, 1985; *Murray* et al., 1985; *Ortiz Monasterio* and *del Campo*, 1985; *Molina* and *Ortiz Monasterio*, 1997; *McCarthy* et al., 2001). Timing, however, still remains an issue to many. The controversy starts regarding aetiology and pathogenesis. According to one theory, damage to the stapedial artery can cause haematoma formation in the first and second branchial arches, resulting in abnormal growth and malformation of the mandible (*Poswillo*, 1973). Others demonstrated that death of neural crest cells can result in dysmorphology of the branchial arches, similar to that found in hemifacial microsomia (*Johnston* and *Bronsky*, 1991).

Growth of the affected mandibular side is another well-debated issue. Some growth studies, meta-analyses and clinical observations have led to the conclusion that the affected mandibular side is not developing at the same rate as the unaffected side (*Poswillo*, 1978; *Murray* et al., 1985; *Kaban* et al., 1998; *Padwa* et al., 1998; *Oeltjen* et al., 2001). Growth and development of the maxilla and midface on the affected side are subsequently hindered and the resulting asymmetry is progressive. Early mandibular reconstruction therefore would then allow maxillary and dento-alveolar development to take place, reducing the need for later surgery. Early repair would also improve body image and socialization of the child.

Other observations and follow-up studies led to the opposite view, i.e., the growth rate of the affected and unaffected sides are equal and the malformation is not progressive with time (*Obwegeser*, 1974; *Rune* et al., 1981, 1983; *Polley* et al., 1997a; *Posnick*, 1998; *Kusnoto* et al., 1999; *Marquez* et al., 2000). Therefore, the majority of the latter authors contend that the mandibular asymmetry should be addressed in adolescence. Intuitively, one would think that intrinsic growth retardation would correlate with the degree of the initial deformity, but this issue also remains unsolved (*Pruzansky*, 1969; *Swanson* and *Murray*, 1978; *Kaban* et al., 1981; *Harvold* et al., 1983; *Polley* et al., 1997; *Drew* et al., 1999).

For those who agree with the progressive nature of facial microsomia, the introduction of distraction osteogenesis in craniomaxillofacial surgery was a major step forward. When compared with costochondral grafting, the advantages of distraction osteogenesis were said to include minimal operative time, low risk of complications, minimal hospitalization time, no donor side morbidity, no need for blood transfusion and no intermaxillary fixation (*McCarthy* et al., 2001). Due to these advantages, patients could be operated on earlier. Early treatment by osteodistraction should have a positive effect on the 'functional matrix', and allow symmetrical growth of the face (*Hollier* et al., 1999b). It was assumed that osteodistraction would produce more stable growth results than does conventional early costochondral or iliac bone grafting (*McCarthy* et al., 1992), but this is not yet proven.

Follow-up studies indicate that the vertical gain is not stable with growth (*Grayson* et al., 1997; *Hollier* et al., 1999b; *Kusnoto* et al., 1999; *Marquez* et al., 2000; *Molina* and *Ortiz Monasterio*, 1995, *Stucki-McCormick* et al., 2001). Furthermore, according to the case report of *Marquez* et al. (2000), the growth rate of the affected mandible may even decrease after osteodistraction. The authors found a long-term effect only at the dentoalveolar level, not at the level of the basal bone. The question is how to differentiate between decreased natural growth and resorption of the regenerated bone. Unfortunately, all follow-up studies longer than 1 year include very few patients (*Swennen* et al., 2001). In addition, the methods used for evaluating the post-distraction growth and stability of the reconstructed vertical ramus are different in each study. In some reports, anthropometric landmarks were used (*Molina* and *Ortiz Monasterio*, 1995), whilst in others different cephalometric analyses were used (orthopantomogram with direct ramus height measurements by *Marquez* et al., 2000), p.a.-cephalogram with measurement of direct–indirect ramus height (*Hollier* et al., 1999b; *Rachmiel* et al., 2001), linear and angular measurements on computerized three-dimensional models from lateral and p.a. cephalograms (*Kusnoto* et al., 1999), orthopantomograms, lateral and p.a. cephalograms without reference to analysis or raw data (*Grayson* et al., 1997). Only one study differentiated between the growth patterns of the ramus and body (*Kusnoto* et al., 1999). The many flaws in the study designs do not allow clear conclusions to be drawn about the vertical stability of the newly generated bone. However, the trend suggests that the results are not better than after early costochondral grafting (*Padwa* et al., 1998). Also, the psychosocial benefit of early surgery for the child is not yet proven. Without question, parents prefer to have a normal-looking child at a young age, but when properly explained, parents will also defer treatment when there is evidence that the end-results are superior. In addition to relapse with growth (necessitating repeated inductive or reconstructive interventions), plus the dubious psychological benefit from an early intervention, there are many technical reasons to defer surgical reconstruction in types I, IIA and even in IIB mandibles until the permanent dentition phase. Orthodontic treatment can then prepare the dental arches in types I and IIA for a conventional three-dimensional orthognathic correction. This means routine intra-oral surgery, occasionally with bone grafts to the maxilla but not to the mandible, and minimal likelyhood for blood transfusions (*Obwegeser*, 1970). The patient compliance required is minimal which seems crucial to the results (*Padwa* et al., 1998). Type IIB mandibles need positional correction, but not necessarily reconstruction of the joint which may result in fibrous ankylosis. Where bone stock is too limited for a large and stable advancement, extra bone volume can be obtained with intra-oral, uni-directional corpus distraction, 1 year prior to the facial rotation surgery. As previously discussed, it remains to be proven that this is superior to conventional interpositional bone grafting.

Only in type III cases is it necessary to reconstruct the temporo-mandibular joint in the mixed dentition phase. These patients suffer from an unstable occlusion and their facial asymmetry is striking. The future glenoid fossa and the mandibular rudiment are hypoplastic and medially displaced. Even with multi-directional distraction osteogenesis, it will be impossible to induce formation of a joint at the right location. Total joint reconstruction (often in combination with orbito-zygomatic reconstruction) is done with costochondral grafting (*Peltomäki*, 1993), cranial bone grafts, lyophilized rib cartilage (*Sailer*, 1976) and/or microvascular fibula transfer. Mandibular symmetry and occlusion can be improved considerably by the costochondral grafting, and the later it is performed, the more stable the results will be (*Padwa* et al., 1998). The final facial rotation surgery is again done in the permanent dentition phase. *Polley* and *Figueroa* (1997) prefer to do 'interim' osteodistraction in type III mandibles to improve, but not to correct, facial symmetry. They do not attempt to reconstruct the lateral temporo-zygomatic deficiency (aesthetics) and the condylar abutment (stable articulation) at that time.

Although in these cases it is theoretically possible to achieve occlusal and facial symmetry with early orthognathic surgery, it is not usually undertaken to avoid harm to the tooth buds when performing a chin osteotomy and Le Fort I-type osteotomy plus the risk of malocclusion later in the permanent dentition. Although it is theoretically possible to avoid tooth buds with early osteodistraction (except in type III mandibles), it should not be proposed as the final solution when full three-dimensional facial and occlusal symmetry cannot be guaranteed. Repeated osteodistraction has already been undertaken frequently (*Polley* and *Figueroa*, 1997; *Hollier* et al., 1999b; *Gosain*, 2001; *McCarthy* et al., 2001) and has shown that mandibular reconstruction is not stable with growth, whether due to relapse or growth impairment. Conventional reconstruction with costochondral grafts in types IIB and III in young patients also requires a second elongation procedure in their late mixed dentition stage (*Kaban* et al., 1998).

To sum up: from this literature review it would appear that:

1. Although the ascending ramus and the affected side is lengthened, mandibular osteodistraction before skeletal maturation in unilateral facial microsomia corrects asymmetry of the mandible for only a relatively short time. Despite over-correction, there is relapse of facial asymmetry. The earlier the correction is undertaken, the more likely it is that osteodistraction procedures will have to be repeated. Whilst over-correction is one remedy, postponing the osteodistraction until the late mixed dentition phase is another.
2. It would appear possible that in some patients the growth of the affected side of the mandible is adversely influenced by the surgeon.
3. The results of osteodistraction in the mandibular body are more stable. In the correction of facial asymmetry, the body of the affected mandible may be more important than the ramus.
4. The superiority of osteodistraction over bone (growth centre) transplantation is not proven.
5. The effect of the osteodistraction on the soft tissue is small and different in each patient (soft-tissue bulk, scars, muscle pull). Tissue expansion will occur only in the direction of the osteodistraction and will not result in lateral augmentation. Soft-tissue management is best carried out separately from skeletal surgery.
6. The frequency of complications such as pin tract infections, cheek abscesses and hypertrophic scars, lead to the conclusion that post-distraction, the soft-tissue condition can be even worse.

CONCLUSION

Future prospective growth studies on osteodistraction are recommended that will:

- differentiate properly between the four Pruzansky–Kaban types of mandibular deformity;
- include adequate numbers in sample groups (multi-centre), treated in a similar way and documented after growth cessation;
- use a standard protocol for three-dimensional evaluation and follow-up (preferably with reproducible computer tomography and quantifying photographic analysis, or otherwise with conventional cephalometric and anthropometrical, and dental model analysis);
- differentiate between vertical and horizontal ramus growth;
- make proper use of the terms 'rate' and 'ratio';
- report on any soft-tissue complications.

Such a prospective three-dimensional growth study of non-operated facial microsomia patients would be of great value for understanding the natural growth potential and to evaluate results of attempts at reconstruction and bone induction.

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