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Mandibular symphyseal distraction osteogenesis: 20 years of experience treating transverse deficiencies with an internal hybrid device

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ABSTRACT

Introduction: Transverse mandibular deficiency has been traditionally managed by orthodontic compensation. However, without resolving the underlying skeletal hypoplasia it leads to high relapse rates. Few studies have reviewed the long-term experience and potential complications of mandibular symphyseal distraction osteogenesis (MSDO) as an alternative treatment method.
Materials and method: A retrospective review of 20 patients (range: 4–19 years; mean: 11.9 years) treated with a hybrid MSDO device over the period of 1996–2017 was conducted. Epidemiological data, medical and dental history as well as photographic and cephalometric analyses were carried out. Furthermore, pre-operative and long-term post-operative status including dental occlusion and tooth-jaw discrepancies were recorded.
Results: The distraction amount ranged from 3 mm to 15 mm (mean: 10 mm) with an average distraction period of 30.9 days. In long-term follow-up (mean: 7.3 years), 14 patients presented a class I and 6 patients presented a class II relationship. Correction of overjet, tooth jaw discrepancy and arch length discrepancy were obtained in 18, 20 and 17 cases respectively. A device malfunction was experienced in 6 patients.
Conclusion: Mandibular widening using a hybrid MSDO device can be efficiently and safely performed in a paediatric population. Further studies confirming our results and analysing facial aesthetic outcomes are warranted.

1. Introduction

Transverse mandibular deficiency is associated with narrow basal and dento-alveolar bone, resulting in anterior dental crowding which is frequently observed in congenital craniofacial malformations (e.g., Treacher-Collins, Crouzon and Silver-Russell syndromes) (Arnaud et al., 2016). The traditional management of arch length and width discrepancies consists of orthodontic treatment including tooth extractions, coronoplasty, vestibular-inclination of the incisors and arch expansion (Lesne et al., 2008; Garreau et al., 2015; Housley et al., 2003). However, it has been shown that orthodontic treatment has limited capability and may lead only to dental compensation without resolving the underlying skeletal hypoplasia, with a high potential of relapse over time. As it severely modifies facial and dental anatomy, this may result in a skeletal class III malocclusion associated with a high rate of relapse of the dental alignment, regardless of whether additional tooth extractions of the maxilla are performed or not (Lesne et al., 2008; Bouletreau and Paulus, 2012; Shields et al., 1985). In the past, vertical symphyseal osteotomies have been performed as the sole

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mode of surgical treatment with rotation of the hemimandibles laterally, either with or without the placement of an additional bone graft (Diner et al., 1997, 1999; Weil et al., 1997; Guerrero et al., 1997). However, this procedure was not well accepted due to its associated mucosal problems, issues with rigid fixation and a high risk of relapse. An alternative strategy to facilitate central incisor eruption and to correct dental crowding and mandibular transverse deficiencies of 7 mm or more in width can be undertaken with the application of mandibular symphyseal distraction osteogenesis (MSDO). In theory, better long-term stability can be achieved with this method as the arch expansion is performed more slowly thus allowing for better adaptation of the soft tissues and the bone regeneration at the osteotomy site. Multiple distraction devices have been described and can be principally divided into bone-borne (Diner et al., 1999) and tooth-borne (Diner et al., 1997) devices with each system having particular advantages and disadvantages (Samchukov et al., 2001; King et al., 2012; Durham et al., 2017; Alkan et al., 2007; Bayram et al., 2007; Carlino et al., 2016). With bone-borne distraction systems, more efficient mandibular basal bone widening can be achieved, resulting in an increased dento-alveolar stability. However, the effectiveness of these devices is disputed because of their potential to cause tooth germ injury. The specific advantages of dental-borne appliances include the lack of the need for surgical removal and transmucosal hardware emergence (Durham et al., 2017; Nadjmi et al., 2015). Disadvantages include an unbalanced amount of arch widening, with more forces applied at the dento-alveolar level and less on the basal bone, which may increase the risk of dental crowding relapse in the long-term. To overcome these problems, Diner et al. (1997) developed a hybrid distraction device with both dental and bone anchorage allowing for the equal distribution of forces at both basal and alveolar bone level. In this study, we describe our hybrid distraction device and present the clinical results and complications observed after 20 years of experience with this procedure.

2. Material and methods

We performed a retrospective analysis of patients treated with a hybrid mandibular symphyseal distraction osteogenesis (MSDO) device. Our study cohort consisted of 20 children (12 males and 8 females) suffering from various craniofacial conditions. The average age at the time of the surgery was 11.9 years old (age range: 4–19 years; SD: 3.4 years). All procedures were carried out at the Department of Maxillofacial and Plastic Surgery, Hôpital Armand-Trousseau and Hôpital Universitaire Necker-Enfants Malades (Paris, France) over the period of 1996–2017. From a chart review, the following information was collected: age, gender, type of congenital malformation, history of medical and orthodontic treatment and surgical protocol (osteotomy site; use of cutting guides or 3D printing models). Furthermore, distraction period, short and long-term complications including distraction failure were noted.

A senior orthodontist carried out a photographic and cephalometric analysis. Pre-operative and long-term post-operative status with regards to dental occlusion, tooth-jaw discrepancies and mouth opening were recorded. The transverse width that was finally achieved was measured on frontal cephalometric by comparing the pre- and postoperative distance between mental nerve foramina.

2.1. Preoperative evaluation and surgical protocol

We routinely perform mandibular symphyseal distraction osteogenesis (MSDO) in cases of congenital mandibular transversal hypoplasia where a transverse widening of 7 mm or more is required and where orthodontic treatment alone is not sufficient to permit the eruption of the central incisors. Pre-operative imaging studies such as computed tomography scans or cone beam CT and 3D printing models are recommended to localize the position of tooth roots and germs and mental nerve foramina but also to define the exact osteotomy sites and distraction device position (Fig. 1A). An occlusal splint is designed and set in place by an orthodontist.

2.2. Device description

The hybrid DO device is an intraoral device, composed of 2 miniplates connected to a central activator. Each miniplate is composed of a lower part with screw holes for bone anchoring and an upper part with screw holes for occlusal splint anchoring (Fig. 1A).

We used a custom-made distractor with a central or lateral activator from the beginning (1996) until 2007, and then the CHOC distractor from 2007 until 2017.

2.3. Surgical procedure and distraction protocol

Patient ventilation is ensured via intra-nasal intubation and surgery is performed through a vestibular approach after infiltration with lidocaine. The osteotomy path is defined by the preoperative set-up and by landmarking of the tooth root prominences. Before carrying out external corticotomies, holes for the future screws are drilled. For the osteotomies, either an oscillating saw or a piezotome is used with or without a cutting guide. Bicortical cutting is performed on the basal bone under and between the roots. A green-stick fracture is then realized on the dento-alveolar bone with a thin osteotome. Mobilization of the osteotomy is done using expansion forceps. The functionality of the inserted distraction device is verified by its activation intraoperative. Finally, the mucosa is closed with interrupted absorbable sutures (Fig. 1B–D).

2.4. Distraction protocol

After a panoramic X-ray has been performed and the activation of the device has been delayed by a latency period of 4 days, distraction was carried out at a rate of 0.5 mm per day. Once the desired transversal widening has been achieved, distractors were left in place for a 3-month consolidation phase before they were removed again.

After DO, patients were treated with conventional multi-ring orthodontics.

3. Results

3.1. Patients

All patients (n = 20) in this study presented with craniofacial conditions including: Brodie syndrome (n = 5), Silver-Russell Syndrome (n = 4), Hanhart syndrome (n = 4), Treacher-Collins syndrome (n = 3), Crouzon syndrome (n = 1), Freedman Sheldon syndrome (n = 1), Cerebro-costo-mandibular syndrome (n = 1) and Pierre Robin sequence (n = 1). The average age at the time of the surgery was 11.9 years (ranging from 4 to 19 years; SD: 3.4 years). Regarding medical and surgical history, we noted prematurity associated with hypotrophy (n = 3), early tracheotomy during infancy (n = 4), enteral nutrition during infancy (n = 4), cleft palate surgery (n = 3) and ramus distraction osteogenesis (n = 1). Pre-operative orthodontic treatment was conducted in 16/20 patients including intermaxillary disjunction (n = 14), vestibular-inclination

Fig. 1. Operative protocol. 1.A: Hybrid MSDO-device on a 3D-printing model. 1.B: Osteotomy guide. 1.C: Completion of osteotomy. 1.D: Distraction device in place.

of the incisors (n = 3), primary tooth extractions (n = 1) and extractions because of tooth decay (n = 1).

3.2. Pre-operative conditions

Orthodontic analysis showed dentoskeletal class malocclusion II in most cases (n = 19/20). All patients presented with a significant overjet (average: 15.4 mm; SD: 5.32, 20-5 mm) (Fig. 3A) and severe tooth jaw and arch-length discrepancy resulting in insufficient alveolar space for canines or incisors to emerge into the right position (Fig. 3B). Four patients presented teeth agenesis and another four patients with a limited mouth opening (average: 23 mm; SD: 3.65 mm), thus complicating the surgical procedure (Table 1).

3.3. Surgical procedure

A vertical median osteotomy was performed in 12/20 patients, a vertical paramedian osteotomy in 7/20 patients and an oblique osteotomy in 1/20 patients. A three-dimensional printing model was used in 6/20 patients and a cutting guide in 2/20 patients.

9 patients were operated using the custom-design distractor (KLS-Martin, Tuttlingen, Germany) and 11 using the CHOC distractor. One patient was operated with the custom-made distractor and underwent new distraction procedure using the new CHOC distractor (CHOC, Montauban, France).

3.4. Postoperative evaluation

The average mandibular widening obtained at the end of our procedure was 8,77 mm (range: 4,2–12 mm; SD: 1,85 mm) with an average distraction period of 30.9 days (range: 15–70 days; SD: 13.1 days). Long-term follow-up (range: 1–20 years; average: 7.3; SD: 5.6) showed dentoskeletal class I in 14/20 patients and class II in 6 cases corrected by orthodontics. One patient presented a midline deviation. The overjet was corrected in 18/20 patients (average: 2.1 mm; SD: 1.2 mm). Tooth jaw discrepancy was corrected in all cases while arch length discrepancy was corrected in 17/20 cases (the 3 remaining patients required vertical branch distraction). Two patients still presented vestibular-inclination of the incisors. No TMJ- or pain-related dysfunction was reported in our study (Table 1, Fig. 2B–D; Fig. 3C, D).

3.5. Complications

No intraoperative complications were noted except for one tooth luxation caused by manipulations during intubation. The distraction protocol was modified in 10/20 cases with an activation performed weekly by the surgeon. During the distraction activation phase, one patient presented pain necessitating device replacement. 4/20 devices malfunctioned and were replaced because of occlusal splint fracture (n = 3), dismantling of the activator body and bad threading of the activating screw (n = 1). Regarding those malfunctions, three happened with the nine custom-made KLS Martin distractors (33.3% malfunction), whereas only one happened with the 12 CHOC distractors used (8.3%).

4. Discussion

In this study, we reviewed our 20-year experience with an internal hybrid mandibular symphyseal distraction osteogenesis (MSDO) device in a large cohort of patients with various craniofacial malformations. First described in 1997 by Diner et al. (1997), this surgical technique has evolved in close cooperation between surgeons and orthodontists to become a standardized procedure in our centre (Diner et al., 1997, 1999; Samchukov et al., 2001). In fact, more than 80% of the patients retrieved a full oral rehabilitation with a correction of the tooth jaw discrepancy, while 13/19 patients with class II malocclusion evolved to a stable class I occlusion. We

Fig. 2. Dental panoramic evolution along with distractor. 2.A: Severe tooth jaw discrepancy with class II malocclusion and significant overjet. 2.B: Severe dental crowding, with insufficient space for incisors to emerge into their right position. 2.C: Resolved tooth jaw discrepancy, with class I occlusion. 2.D: Resolved dental crowding, with all teeth in their correct position from 47 to 37.

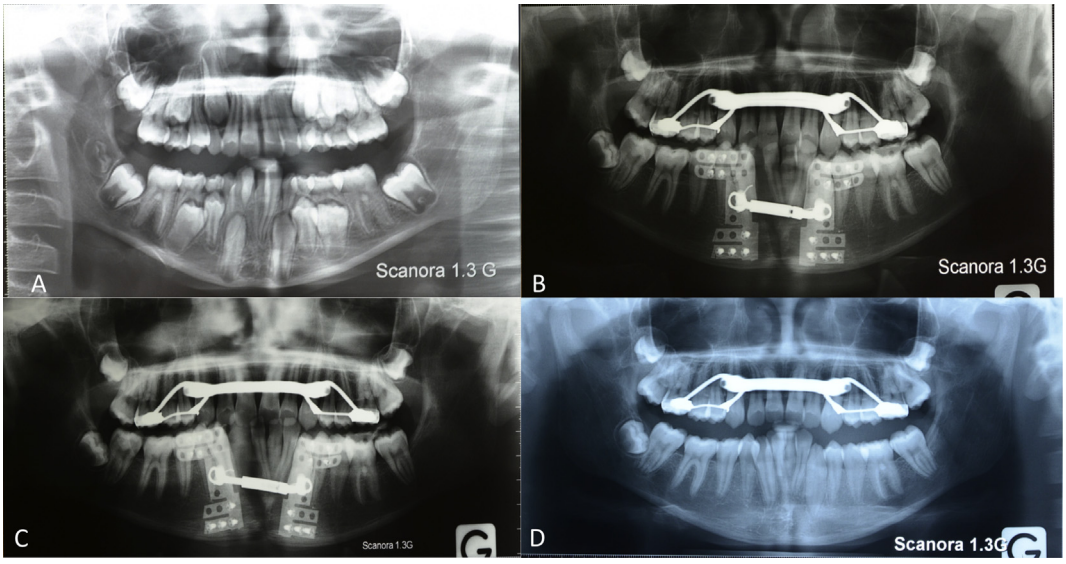

Fig. 3. Oral and dental photographs before (A, B) and after (C, D) mandibular symphyseal distraction osteogenesis. 3.A: Severe tooth jaw discrepancy with class II malocclusion and significant overjet. 3.B: Severe dental crowding, with insufficient space for incisors to emerge into their right position. 3.C: Resolved tooth jaw discrepancy, with class I occlusion. 3.D: Resolved dental crowding, with all teeth in their correct position from 47 to 37.

hypothesize that this correction is due to a condyle rotation, initiated by distraction osteogenesis, which moved the vertical mandibular branch in a valgus direction.

When orthodontic treatment is performed alone, high relapse rates, with major periodontal adverse effects, have been frequently described in the literature. Blake and Bibby (1998) found a relapse rate of more than 30% following retention removal, similar to Housley et al. (2003), who reported a relapse rate of 20–30%.

In contrast to orthodontic compensation alone, the application of mandibular symphyseal distraction osteogenesis (MSDO) has been shown to pose a lower risk of relapse. One of the reasons may be that the produced bone regenerate adds more stability to the innate mandibular basal bone level. When analyzing our cohort, we did not observe any relapse after treatment with MSDO, even after long-term follow-up (range: 1–20 years; average: 7.3 years). No relapse had been shown even in patients without intrinsic mandibular growth, as Treacher Collins patients, confirming the stability of the procedure.

However, the threshold at which orthodontic treatment does not seem to be feasible as the sole mode of treatment of transverse

Table 1
Orthodontic status before and after long-term follow-up (SD = standard deviation, N/A = non applicable).

		Before (n = 20)	After (n = 20)
Follow-up in years (average/SD)		N/A	7.3/5.6
Dento-skeletal class	I	1	14
	II	19	6
Arch length discrepancy		20	3
Tooth jaw discrepancy	Mandible	20	0
	Maxilla	20	0
Overjet in mm (average/SD)		15.4/3.6	2.1/1.2
Patient with mouth opening limitation		4	4

mandibular deficiency is still a subject of debate. While Garreau et al. (2015) propose that a mandibular widening of over 3 mm cannot be compensated by orthodontic treatment alone and requires MSDO, other authors recommend a higher cut-off value of 7 mm also taking the risk for potential surgical side-effects into account (Winsauer et al., 2017; Alkan et al., 2007). However, these values have only been discussed for the treatment of adults. From our long-term experience in the paediatric population, we also advocate a conservative approach and apply mandibular symphyseal distraction osteogenesis (MSDO) for transverse deficiencies above 7 mm.

Although aesthetic outcomes were not specifically analysed in this study, their beneficial effects on facial dysmorphism appear to be obvious. For example, a hypoplastic narrow chin can be frequently encountered in Freeman and Silver-Russel syndromes with a characteristic “whistling-face”. With the application of MSDO, an acceptable chin width could be restored in these cases. However, studies confirming our observations on the aesthetic and facial effects of mandibular widening procedures in a paediatric population are still lacking.

Furthermore, there is a lot of controversy in the literature concerning the advantages and disadvantages of bone-borne, tooth-borne and hybrid-borne devices and experiences vary from study to study (King et al., 2012; Uckan et al., 2006; Nadjmi et al., 2015; Alkan et al., 2007; Bayram et al., 2007; Guerrero et al., 1997). In their pioneering investigation, Guerrero et al., (1997) found that tooth-borne Hyrax appliances caused disproportionate movement of teeth and bone both experimentally and clinically and were therefore inferior to bone-borne osteo-distractors. In a more recent study, Alkan et al. (2007) compared all three types of devices (tooth-borne, bone-borne and hybrid) and concluded that a lingually placed tooth-borne Hyrax appliance is preferred because of its minimal invasiveness and comfort for the patient and surgeon. However, in cases in which the space for the placement of a Hyrax appliance in the lingual region would be insufficient, they advocated the use of hybrid distractors as an alternative. From the literature, it appears that bone-borne devices have a higher risk of hardware failure such as device breakage (Alkan et al., 2007). In our surgical protocol, a total vertical midline osteotomy of the mandibular symphysis is performed and the hybrid distraction device is fixed to both bone and teeth (double splint and basal bone fixation). In theory, this permits forces to be equally applied over the entire height of the mandible (alveolar and basal bone). Moreover, such a device allows the surgeon to change the activation screw when required to increase its widening capacity over time, given the initial dental crowding. In our cohort, we did not observe any discrepancy between basal and alveolar bone widening, however we initially also noted a high rate of device malfunction (30 %). Modifications which were made to our hybrid distractor, first designed and created in a custom-made and then in an industrial fashion because of a high malfunction rate, resulted in an improvement in safety and efficiency, and no further device failure was observed in patients operated on after 2010. Problems related to the mandibular osteotomy, such as wound dehiscence or mental nerve irritation have been reported in the literature, however symptoms usually appear to be transient, not necessitating any further treatment (Kewitt and Van Sickels, 1999; Raoul et al., 2009). Other complications related to the use of tooth-borne and bone-borne distractors include impaired tooth vitality, root exposure, and deep pockets (Uckan et al., 2006; von Bremen et al., 2008; Kewitt and Van Sickels, 1999). However, none of these complications was observed in our study cohort. We believe that the use of a stereolithographic mandible model, which permits the surgeon to pre-operatively landmark tooth roots and mental nerve foramina and plan the osteotomy, significantly reduces the risk for tooth or neurosensory related damage.

This surgery, in growing children, is considered as interceptive surgery, to accompany children during their development. Obviously, it does not exclude end-of-growth orthognathic surgery. This should be explained to the child and parents.

Finally, mandibular symphyseal distraction osteogenesis (MSDO) is a safe procedure that may be performed under local or general anaesthetic. Multiple studies suggest that MSDO can be carried out under local anaesthetic with or without associated intravenous sedation. However, with regards to paediatric patients, we consider general anaesthesia to be mandatory for both the surgeon's and patient's compliance and comfort.

5. Conclusion

In the present study, we demonstrated the efficacy and safety of mandibular symphyseal distraction osteogenesis using a hybrid device in a large paediatric cohort. Patients with transverse mandibular hypoplasia of over 7 mm and tooth-jaw discrepancies could be successfully treated with no signs of relapse in the long-term follow-up. Future studies on MSDO in a paediatric population are however required to confirm our results and should also include a detailed analysis of facial aesthetic outcomes.

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Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.jcms.2019.01.001.

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