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Mandibular distraction osteogenesis in children with Pierre Robin sequence: long-term analysis of teeth and jaw growth

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Abstract

Treatment of children with Pierre Robin sequence (PRS) having a hypoplastic mandible and upper airway distress after birth may consist of external distraction devices. Shape anomalies of the permanent molars and positional changes due to surgery have been documented. The aim of this study is to compare the long-term effects (>5 years) on the growth pattern of PRS-patients treated with an external mandibular distraction device with no-surgery cases and to investigate the dental development or damage. A retrospective cohort study was performed. PRS-patients with and without surgery were included. A digital cephalometric analysis was made to evaluate the growth pattern of the mandible between groups as well as with normal values. Nine of 19 patients underwent an external mandibular distraction. All children were extubated after 4-5 days with no signs of respiratory distress. Screw and device loosening presented in one patient. The articular and sellar angles were significantly larger and smaller, respectively, in the Surgery group. Mandibular distraction surgery might result in a 'growth boost' compared to the No-surgery group. No significant difference in dental development was found. Mandibular distraction osteogenesis is an effective way of relieving severe upper airway obstruction.

Keywords: Mandible; Distraction; Pierre Robin; Jaw Growth

Introduction

Pierre Robin Sequence (PRS) is characterised by micrognathia, glossoptosis, and upper airway obstruction (UAO) often in combination with a U-shaped cleft palate.[1] The incidence ranges between 1 in 8 000 births[2] to 1 in 30 000 births.[3]

Upper airway obstruction may impair breathing, feeding, and, consequently, growth. Most PRS-patients can be treated conservatively with nonsurgical measures, for example a nasopharyngeal airway or continuous positive airway pressure (CPAP).[4,5] Alternative treatment options are mandibular traction devices[6] or tongue-lip adhesion,[7] with the aim of keeping the airway sufficiently open.[8,9] In advanced cases, the airway obstruction can be life-threatening. Tracheostomy is an effective procedure to address this problem.[10] On average, the age at decannulation is 3.1 years, subjecting PRS-patients to a long time of potential problems.[3] Because of the high morbidity, mortality and costs associated with this invasive measure, other alternatives have been investigated.[11–13]

Since the introduction of mandibular distraction osteogenesis (MDO) in 1995, this treatment has proven to be very

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effective, achieving 97.6% decannulation in PRS-patients.[14] Mandibular distraction devices require an osteotomy but allow distraction of the mandible while protecting the temporomandibular joint. The upper airway size at the retropalatal level will increase, leading to physiological improvements in the apnoea-hypopnea index and minimum oxygen saturation.[15]

Short- and mid-term effects of MDO on jaw growth have been described in various articles.[16–19] Shape anomalies of the permanent molars, root changes, and positional changes due to the surgery have also been documented.[20]

The aim of this study is to compare the long-term effects (>5 years) on the growth pattern of the mandible of PRS-patients treated with an external mandibular distraction device with no-surgery cases and to investigate the dental development or damage.

Material and methods

This study was approved by the ethical board of the University Hospital of Antwerp (B3002020000155). Informed consent was obtained from all representatives of the included patients.

Patient population

Inclusion criteria were patients with isolated PRS. A retrospective cohort study was performed and all included patients had follow up of at least five years and were born between 1996 and 2014. The patients were treated by the same maxillofacial surgeon.

PRS-patients with follow up of less than five years or PRS-patients who also had a combination of neuromuscular diseases or syndromic patients were excluded.

Surgery was preferred if patients had clinical obstruction symptoms as a consequence of PRS that could not be managed with conservative measures. Before MDO, all children with an indication for surgery had a polysomnography (PSG), microlaryngoscopy and bronchoscopy (MLB), which showed moderate to severe obstructive sleep apnoea (OSA).[21]

All patients had a cleft palate, which was surgically corrected at the age of nine months (soft palate) and 18 months (hard palate) according to the cleft treatment protocol of our institution.[22]

The patient records were screened retrospectively and reviewed to evaluate long-term outcomes. All included patients were recalled for a physical examination. An orthopantomogram and lateral cephalogram (Planmeca ProMax 2DS2, 68kV, 10mA; Planmeca) were made for evaluation of dental development and skeletal analysis.

Treatment plan and surgical technique

Patients in whom conservative measures would not suffice and a tracheostomy was considered, underwent MDO with a Molina unidirectional external mandibular distractor (KLS Martin Group) as their primary surgical therapy. All patients received an endoscopy by the ENT surgeon prior to MDO to exclude any other cause of airway obstruction.

No preoperative 3D planning was performed, with the 'as low as reasonably achievable' (ALARA) principle in mind. Placing guides would mean more degloving of the mandible and therefore more swelling.

After an intraoral posterior vestibular incision, subperiosteal dissection was performed. Bilaterally, a minimal invasive oblique corticotomy was made from the retromolar region towards the mandibular angle, using a small reciprocating saw or a piezotome. The corticotomy was performed as far as possible in the lingual side while protecting the overlying soft tissue. The percutaneous distraction pins were placed posteriorly and anteriorly to the corticotomy, a few millimetres above the mandibular border, while extra skin was pinched between the pins to avoid traction force on the skin during the distraction phase (Fig. 1). The division between the proximal and distal segments was achieved by digital pressure on the distal segment. Subsequently external distractors were placed over the pins and the activation test was performed.

Perioperatively, the overjet was measured between the upper and lower alveolar ridge to calculate the amount of distraction needed.

The distraction device was activated immediately, at a rate of 2 × 1 mm each day. The distraction was continued until a neutral-relation was achieved between the upper and lower jaw. No overcorrection was done, according to the surgeon's preference. The distraction pins were removed during the check-up consultation after a consolidation phase of minimum four weeks.

Clinical course

Extubation was possible after 4-5 days (8-10 mm distraction), after careful clinical evaluation by cardiorespiratory monitoring and approval of the intensive care specialist.

Postoperatively, feeding started directly by a nasogastric tube and oral feeding was started within two weeks after the surgery. A Habermann feeder was used in presence of a cleft palate.

Data collection

Primary outcomes consisted of mandibular growth measured by cephalometric landmarks (Fig. 2). Dental development and damage related to the surgery in the premolar-molar region of the mandible, clinically presenting as agenesis, crown shape changes, root malformations or positional changes were investigated. Percentages were calculated on ten teeth per case. An analysis between the two treatment groups was made as well as a comparison to normal values of a healthy population.

A digital cephalometry in OnyxCeph (Image Instruments) was performed based on the Bjork-Jarabak skeletal analysis and landmarks used in the study published by Paes et al.[17]

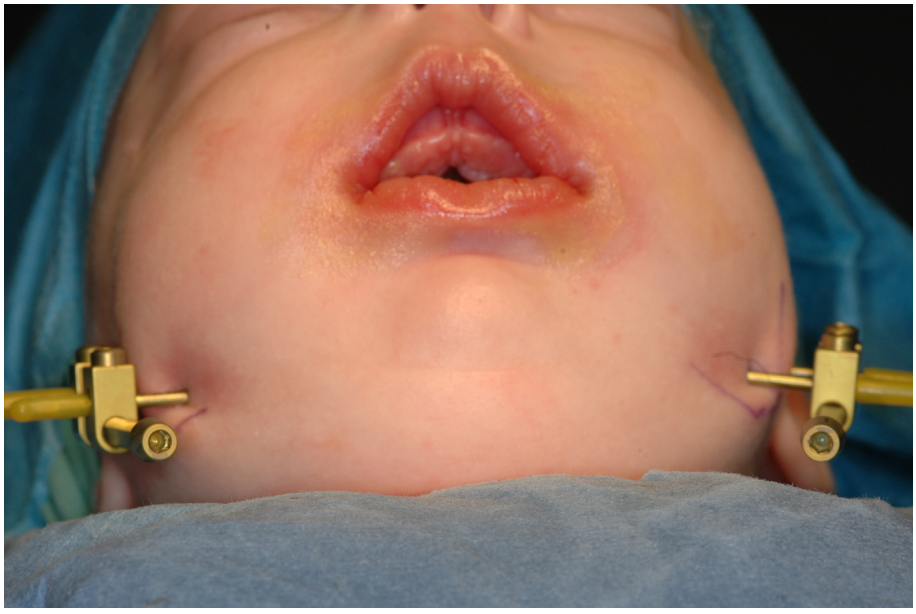

Fig. 1. The percutaneous distraction pins were placed posteriorly and anteriorly to the corticotomy, a few millimetres above the mandibular border, while extra skin was pinched between the pins. This was to avoid traction force on the skin during the distraction phase.

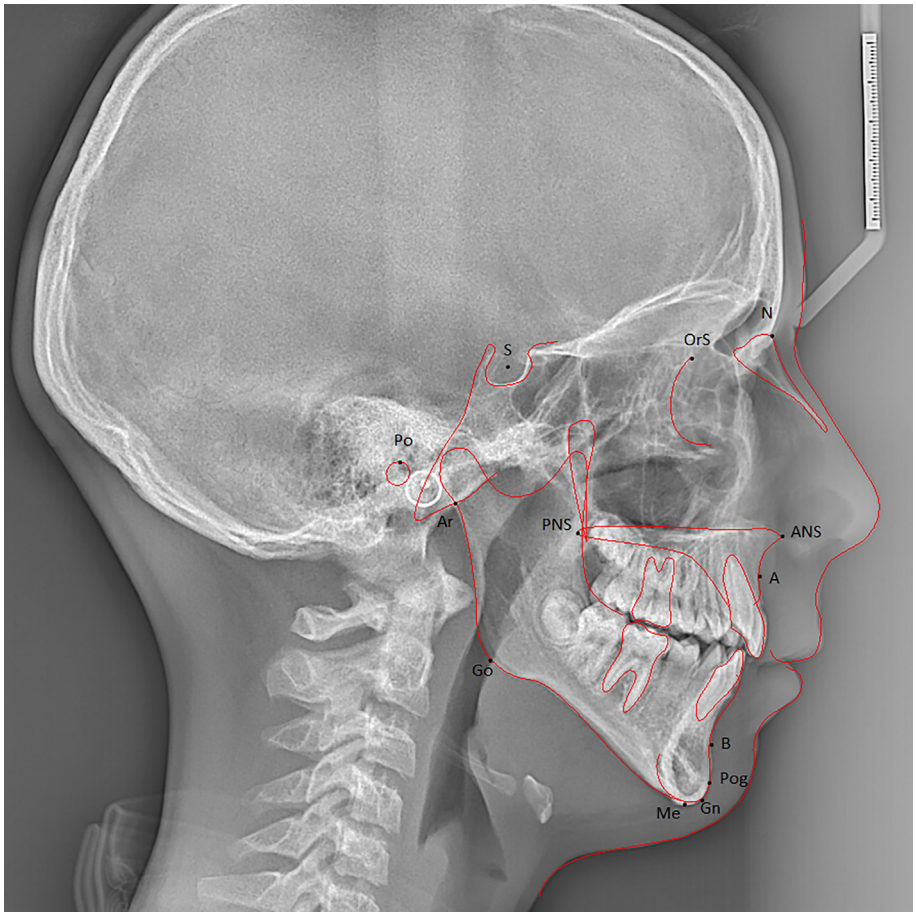

Fig. 2. Cephalometric analysis: Ar (articulare), S (sella), Ors (orbitale superior), Or (Orbitale), ANS (spina nasalis anterior), PNS (spina nasalis posterior), A (A-point), B (B-point), Pog (pogonion), Me (Menton), Go (gonion), Gn (gnathion), Po (porion). Anatomical lines automatically drawn by the software (Onyx) based on the landmark placement.

The articular angle (S-Ar-Go), sellar angle (N-S-Ar) and gonial angle (Ar-Go-Me) are used in the analysis of facial growth patterns. The sum of the articular angle, sellar angle, and gonial angle is called the Bjork sum angle and will be larger and smaller than 396° in an open (clockwise) and closed (counter clockwise) growth pattern, respectively.[23–25]

The age- and gender-specific cephalometric data of the University of Bonn were used as reference values for the normal population.[26]

Statistical analysis

The data were collected and statistical analysis was performed with JMP software (SAS Institute Inc).

Significant differences in dental status between the two groups were evaluated using the Fisher's exact test for small samples.

The cephalometric analysis is presented for the total population as well as for the two treatment groups. The measurement data was tested for normality using the Shapiro-Wilk test. Statistical differences were analysed using a two-tailed independent samples *t* test for normal distributed data and using a Wilcoxon rank-sum test for non-normal distributed data.

To compare the cephalometric measurements with normal values, a one-sample *t* test assuming a mean value of zero for the z-scores was performed in order to find statistically significant differences between the two groups and the normal population.

Results

A total of 43 PRS-patients presented to our department between 1996 and 2014. Ten patients were lost to follow up or had insufficient data to be included. Eleven patients were excluded because of certain syndromes or neuromuscular diseases. Three patients with a follow up of less than five years were also excluded.

A total of 19 patients was included of whom nine underwent MDO (Surgery group, n = 9) and 10 patients (No-surgery group, n = 10) had a conservative follow up. The median age at surgery for the Surgery group was nine weeks. The average was 21 weeks due to an outlier. The demographics are shown in Table 1.

Short-term complications of surgery included device loosening in one patient (11%). A small procedure under sedative anaesthesia was needed to replace the device. One patient had a local infection on the pin opening, for which topical and oral antibiotics were administered.

Dental analysis

Statistical analysis of the dental status in the surgery and no-surgery group did not show any significant differences (Table 2).

In the Surgery group, four patients (44%) had agenesia in the lower jaw. In the No-surgery group, there were six patients (60%). The second premolars were the most often agenetic: three patients in the Surgery group and five patients in the No-surgery group, respectively.

Regarding shape changes of the crown, there were only two patients in the Surgery group. Shape changes included hypoplasia and in one tooth the presence of a hole at the enamel/dentin junction (Fig. 3).

Positional changes were present in two patients in the surgery group, affecting two of the three teeth that also showed shape changes. Root malformations were present in one case in the Surgery group, affecting both first permanent molars.

Cephalometric analysis

Table 3 shows the cephalometric values of the Surgery and No-surgery groups compared with normal values as well as intergroup comparison.

The SNA-angle, which represents maxillary growth, was not significantly smaller in the Surgery group compared to the No-surgery group. But compared with normal values, the SNA-angle for the surgery group was significantly smaller ($p = 0.0385$).

There were no significant differences in the intergroup comparison in any of the measurements besides the articular angle and the sellar angle. The articular and sellar angle was significantly larger and smaller in the Surgery group, respectively. The gonial angle showed no significant difference.

Table 1
Demographic characteristics of the Surgery and No surgery groups. P values < 0.05 are considered to be statistically significant (M = Male, F = Female).

Variable	Total population	Surgery	No surgery	p value
Number of patients	19	9	10	
Gender [No. (%)]	M: 7 (37%)	M: 6 (67%)	M: 1 (10%)	Pearson
	F: 12 (63%)	F: 3 (33%)	F: 9 (90%)	0.0106 *
Age at tracing [mean (SD)] and/or median [range] (years)	13 (4)	13 (4)	13 (4)	*t* test
	Median = 13	13 [7–19]	14 [6–18]	0.9952
Age at surgery [mean (SD)] (weeks)	/	21.6 (38.7)	/	
		Median = 9.0		
Follow-up time (years)	13 (4)	12 (4)	13 (4)	*t* test
	Median = 13	Median = 11	Median = 14	0.6829

Table 2
Dental analysis on the panoramic radiograph. Data are No. (%).

Variables	Surgery (cases n = 9)	No Surgery (cases n = 10)	Fisher's exact test p value
Agenesia:			
Cases	4 (44%)	6 (60%)	0.6563
Affected teeth:	5 (5%)	12 (12%)	0.2091
Cases with 1 affected tooth	3	2	
Cases with 2 affected teeth	1	3	
Cases with 3 affected teeth	0	1	
Canines:	0	2	
Unilateral		0	
Bilateral		1	
First premolar (34, 44)	0	0	
Second premolar (35, 45)	4	9	
Unilateral	2	1	
Bilateral	1	4	
First molar (36, 46)	0	0	
Second molar (37, 47)	1	1	
Unilateral	1	1	
Bilateral	0	0	
Shape changes crown:			
Cases	2 (22%)	0	0.2105
Affected teeth	3(3%)	0	0.1044
Case 1	36	/	
Case 2	36, 45	/	
Positional changes:			
Cases	2 (22%)	0	0.2105
Affected teeth	2 (2%)	0	0.2231
Case 1	36	/	
Case 2	45	/	
Root malformation:			
Cases	1(11%)	0	0.4737
Affected teeth	2 (2%)	0	0.2231
Affected teeth	36, 46		

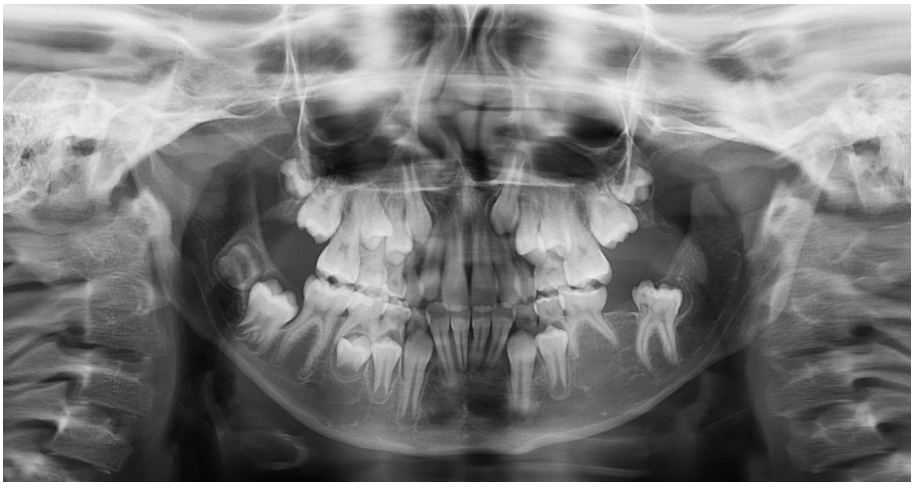

Fig. 3. Panoramic radiograph: positional change of element 36: more distally positioned with also a crown malformation present at the mesial side of this tooth. Agenesia of tooth 35.

The Bjork sum angle was 400.7° and 396.7° for the Surgery and No-surgery group, respectively, and showed no significant difference. An example of a comparison of the growth pattern in both groups can be seen in supplementary Figure 4 (online only). Other indicators for a more vertical clockwise growth pattern did not differ significantly.

The total mandibular length (Ar-Pg) and the vertical component ramus height (Ar-Go) in the Surgery group were significantly smaller compared to normal values. The horizontal component Go-Pg and Go-Me is significantly shorter for all PRS-patients, regardless of their treatment. There was no significant difference in the intergroup comparison.

Table 3
Cephalometric values of the Surgery and No Surgery groups. Data are mean (SD) or p values.

Variable (°)	Surgery,	Surgery vs normal values	No surgery,	No surgery vs normal values	Surgery vs No surgery
SNA	75.7 (6.2)	0.0385*	78.5 (5.6)	0.3547	0.321
SNB	74.9 (8.2)	0.2986	77.3 (5.7)	0.5524	0.483
SNPg	75.5 (8.4)	0.3453	78.4 (5.5)	0.8969	0.398
SN–GoGn	38.8 (9.5)	Variable	33.6 (7.3)	0.2847	0.206
SN–GoMe	41.1 (10.3)	0.1184	36.8 (7.1)	0.5304	0.317
PP–MP	30.7 (10.0)	0.6667	29.3 (8.5)	0.3223†	0.737
PP–GoGn	28.1 (9.2)	0.4352	25.9 (8.7)	0.9154	0.598
Ar–Go–Gn	134.3 (12.9)	0.4122	135.0 (6.2)	0.0544	0.890
N–S–Ar	117.7 (10.7)	0.1125	128.1 (6.8)	0.1759	0.0254*
Sellar angle					
S–Ar–Go	148.7 (12.5)	0.2103	133.6 (10.5)	0.0200*	0.012*
Articular angle					
Ar–Go–Me	134.3 (12.9)	0.4177	135.0 (6.1)	0.0502	0.879
onial angle					
Bjork sum angle	400.6 (10.9)	0.2353	396.7 (7.1)	0.7416	0.3779
Ar–Pg (mm),	90.4 (10.5) median = 84.9	0.0224*	97.8 (11.8)median = 99.0	0.6250†	0.206†
mandibular length					
Ar–Go (mm)	38.5 (11.7)	0.0444*	44.5 (7.6)	0.1275	0.207
mandibular ramus height					
Go–Me (mm)	59.8 (5.3)	0.0002*	62.4 (6.7)	0.0027*	0.358
Go–Pg (mm)	61.6 (5.5)	0.0003* †	63.7 (5.9)	< 0.0001*	0.429
S–Go (mm)	67.0 (11.4)	0.6465	67.8 (8.3)	0.6804	0.867
posterior facial height					
N–Me (mm)	110.0 (9.3)	0.5738	107.5 (13.8)	0.8972	0.647
anterior facial height					

SNA, angle between sella, nasion and A point; *SNB*, angle between sella, nasion and B point; *SNPg*, angle between sella, nasion and pogonion; *Pg-N-B*, angle between pogonion, nasion and B point; *SN-GoGn*, angle between sella-nasion and gonion-gnathion; *SN-GoMe*, angle between sella-nasion and gonion-menton; *PP-MP*, angle between palatal plane and mandibular plane; *PP-GoGn*, angle between palatal plane and gonion-gnathion; *Go-Me*, distance from gonion to menton; *Ar-Go* (mandibular ramus height), distance from articulare to gonion; *Ar-Go-Gn*, angle between articulare, gonion and gnathion; *N-S-Ar (sellar/sellar angle),* angle between the nasion, sella and articulare; *S-Ar-Go (articular angle),* angle between sella, articulare and gonion; *Ar-Go-Me (gonial angle)*, angle between articulare, gonion and menton; *Go-Pg*, distance between gonion and pogonion; *Ar-Pg (mandibular length)*, distance between articulare and pogonion; *Go-Gn*, distance between gonion and gnathion; *Se-N*, distance between sella and nasion; *S-Go (posterior facial height)*, distance between sella and gonion; *N-Me (anterior facial height)*, distance between nasion and menton.

* p value < 0.05, statistical significance.

† Wilcoxon test performed instead of *t* test.

The anterior facial height was not significantly larger in the Surgery group versus the No surgery group. The posterior facial height was comparable for both groups. Compared with the normal population, there was no significant difference found in both anterior and posterior facial height in the Surgery and No-surgery group.

The long-term clinical outcome at age 15 of one of the patients in the surgery group is shown in supplementary Figure 5 (online only). No further mandibular advancement techniques were necessary.

Discussion

The aim of this study is to evaluate the long-term effects (>5 years) on the growth pattern of the mandible in infants treated with MDO and to investigate the dental development or damage related to its technique.

Regarding jaw growth, there was no significant difference. One can question whether the patients who had MDO might have had a more retrusive jaw at birth. If this is the case, one can interpret that MDO results in a 'growth boost' so the final jaw length is comparable to the No-surgery group.

Regarding maxillary growth, the SNA value is only significantly smaller in the Surgery group compared to normal values. This restricted maxillary growth can be explained by the presence of a cleft palate and the effect of surgical intervention.

Many articles suggest a more pronounced vertical growth pattern associated with PRS.[17,27] However, this could not be proven in our study, as some parameters indicated that more vertical growth was not significantly larger. The limited data can be the causative factor.

Dental trauma has often been described in literature as a consequence of the osteotomy or pin placement. The use of external distractors is considered to be a risk factor for injuring dental buds because of the bicortical pin placement.[28] Alternative osteotomy-designs (inverted L-shape) and using internal distractors have been proposed to avoid injuring tooth buds during the osteotomy.[16]

No significant differences were found in dental injury or shape/root malformations, but they concerned only those patients who had had MDO. The absence of significance is presumably due to the small sample size. The second premolars were most often missing, which are also the most common congenitally missing teeth. Futhermore, the smaller mandible in PRS-patients has the potential to affect tooth development, since there is an association between mandibular morphology and dental agenesis.[29]

Damage to teeth may be induced by bicortical pin placement. This is a major drawback for the use of external distractors. However, this kind of damage was only seen in two patients. The osteotomy design is of importance in preventing tooth damage. In this study, the surgeon performed an oblique corticotomy at the angle of the mandible.

One study reporting long-term outcomes after internal resorbable devices showed 70 percent positional/directional changes in molars and another 70% of cases with shape anomalies.[17] A study of 34 patients with a mean age of 8.1 years during MDO showed that almost half of the patients did not have any tooth alterations after MDO, and more than 20% presented only bud distalisation.[20] However, this patient population is not comparable with the patients treated with distraction devices in this study, since the median age during surgery is nine weeks in our population. The stage of dental development is therefore very different. Another retrospective study including 141 patients over a time frame of 16 years reported only 0.67% damage to tooth follicles or teeth.[30] It is obvious that there is a broad variation in reports of dental damage due to the differences in surgical techniques, devices, age at which the procedure is done, and the definition of tooth damage.

This study has potential limitations, including the small size of patient groups and the retrospective nature of this study. In future, having a multicentric prospective study with a large patient population, might give more insight on this interesting topic.

Conclusion

There is paucity in literature regarding the long-term effects of MDO on the mandibular growth and dental damage in PRS-patients. Negative effects on growth of the mandible or a more vertical growth pattern could not be seen in this study. However, the intrinsic growth potential of PRS-patients seems to be normal, which does not exclude subsequent procedures for correction of the jaw relation at an older age. Dental damage is rare if the osteotomy and pins are placed strategically and should not be used as a drawback. MDO is a valuable, safe treatment for PRS-patients where a tracheostomy or long-term CPAP treatment is considered.

Conflict of interest

We have no conflicts of interest.

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Ethics statement/confirmation of patient permission

This study was approved by the ethical board of the University Hospital of Antwerp (B3002020000155). Informed consent was obtained from all representatives of the included patients.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.bjoms.2024.04.008.

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