OPEN

PRS GLOBAL OPEN

ORIGINAL ARTICLE

Pediatric/Craniofacial

Infant Mandibular Distraction for Upper Airway Obstruction: A Clinical Audit

Ashim N. Adhikari, MD, BDSc, FRACDS,*
Andrew A.C. Heggie, MBBS, MDSc, FRACDS(OMS),†,‡
Jocelyn M. Shand, MBBS, BDS, FRACDS(OMS),‡
Patrishia Bordbar, MBBS, BDSc, FRACDS(OMS),‡
Anastasia Pellicano, MBBS, FRACP,§
Nicky Kilpatrick, BDS, PhD, FRACDS(Paeds)†,¶

Background: Mandibular distraction osteogenesis (MDO) is an effective method of treating upper airway obstruction (UAO) in micrognathic infants. The short-term outcomes include relief of UAO, avoidance of tracheostomy, and prompt discharge from hospital. However, it is a significant surgical procedure with potential associated morbidities. This study describes a cohort of infants managed using MDO over a twelve-year period.

Methods: A retrospective chart review was undertaken for children who had MDO before the age of 5 years between 2000 and 2012. This was followed by a clinical review of the same cohort specifically looking for dental anomalies, nerve injuries, and scar cosmesis.

Results: Seventy-three children underwent MDO at a mean age of 2 months [interquartile range (IQR), 1.7–4.2] for nonsyndromic infants and 3.3 months (IQR, 2.1–7.4) for those with syndromes. Infants were discharged from hospital, on average, 15 days after procedure. After MDO, of the 9 who were previously tracheostomy dependent, 5 (56%) were decannulated within 12 months and none of the nontracheostomy-dependent children required further airway assistance. The majority of children required supplemental feeding preoperatively but, 12 months postoperatively, 97% of the nonsyndromic infants fed orally. Thirty-nine children (53%) were reviewed clinically [median age, 5.1 y (IQR, 3.9–6.5)] with 18 being syndromic. Many of the mandibular first permanent and second primary molars had developmental defects, but there was a low rate of neurosensory deficit and good scar cosmesis.

Conclusions: This study contributes further to the evidence base underpinning the management of micrognathic infants with UAO. *(Plast Reconstr Surg Glob Open 2016;4:e812; doi: 10.1097/GOX.0000000000000822; Published online 20 July 2016.)*

Micrognathia has been described as a feature of many syndromes[1] and may be associated with upper airway obstruction (UAO) and occurs in infants with craniofacial anomalies such as Pierre Robin sequence, Treacher Collins syndrome, and craniofacial microsomia variants. In these anomalies, the hypoplastic mandible is retropositioned causing posterior displacement of the tongue (glossoptosis) and a concomitant reduction of the oropharyngeal airway that leads to UAO. Robin sequence is the commonest of these anomalies,[2] with an estimated incidence of 1:8,500–14,000.[3] Infants with severe airway obstruction suffer hypoxia that may result in respiratory failure, cardiovascular complications, and feeding difficulties that may be fatal.

There is currently no evidence-based consensus regarding the best management for patients with moderate-to-severe UAO.[4] Nonsurgical options include the use of a nasopharyngeal airway,[5–7] continuous positive airway pressure,[8,9] and modified infant feeding plates.[10] Surgical options have included tongue–lip adhesion (TLA)[11] and tracheostomy.[12] Neither procedures are ideal; TLA is rarely a definitive procedure[13] and tracheostomy, although a complete cure for UAO, is associated with significant morbidity and poses a heavy burden on families, making it a treatment of "last resort."[14,15]

*From the *Faculty of Medicine, Dentistry and Health Sciences and †Department of Paediatrics, University of Melbourne, Melbourne, Victoria, Australia; ‡Head, Oral & Maxillofacial Surgery Unit, Department of Plastic and Maxillofacial Surgery, the Royal Children's Hospital, Melbourne, Victoria, Australia; §Department of Neonatology, Royal Children's Hospital, Melbourne, Victoria, Australia; and ¶Plastic Surgery Research, Murdoch Childrens Research Institute, the Royal Children's Hospital, Melbourne, Victoria, Australia.*

Received for publication 02 February, 2016; accepted 20 May, 2016.

DOI: 10.1097/GOX.0000000000000822

Disclosure: *The authors have no financial interest to declare in relation to the content of this article. The Article Processing Charge was paid for by the authors.*

Mandibular distraction osteogenesis (MDO) has been introduced as an effective method of treating UAO related to micrognathia in infants. This surgical option has a low morbidity, few short-term complications,[12,16,17] and superior outcomes to TLA.[18] However, as an emerging technique, the literature consists of short-term follow-up of small cohorts[19] with few studies describing the longer-term effects of MDO, particularly on the developing mandible and dentition.[20,21]

The aim of this study was to review a series of micrognathic infants with UAO that have undergone MDO at the Royal Children's Hospital (RCH). The study was conducted in 2 phases: a retrospective chart review of the first-year post-MDO with respect to airway, feeding, growth, hospital stay, and short-term complications followed by a clinical review of these same patients to assess the longer-term effects on dental development, inferior alveolar nerve function, and scar cosmesis.

PATIENTS AND METHODS

Approval for the study was granted by the RCH Human Research Ethics Committee (HREC 33253). All patients who underwent MDO under the age of 5 years between January 2000 and December 2012 at the RCH were identified from the Oral and Maxillofacial Surgery Unit's database. All patients had been operated on by 1 of the 2 surgeons (A.A.C.H. and J.M.S.) using a standardized surgical technique that has been described previously.[22] After general anesthesia and orotracheal intubation, local anesthetic solution was infiltrated, and a 2.5-cm submandibular incision was made 1.5 cm below the lower border of the mandible. A dissection to the lower border of the mandible was made deep to the cervical fascia to expose the ramus and posterior body of the mandible. The distraction device was then inserted into the desired position and the activation arm directed posteriorly exiting percutaneously beneath the ear lobe. The proposed osteotomy was made with a fissure burr from the retromolar region in an arc posterior to tooth buds and carried to just anterior to the angle of the mandible. The device (Zurich Paediatric Ramus Distractor, KLS Martin, Tuttlingen, Germany) was then attached with self-tapping 1.5-mm diameter, 5- to 7-mm long bone screws to the lateral cortex. The wound was then closed in layers with resorbable sutures and dressings applied. An identical procedure was then performed on the contralateral side.

The patients remained intubated and were returned to the neonatal intensive care unit where they remained sedated. The devices were activated at a rate of 1.5 mm per day (activated 0.5 mm 3 times daily) to a maximum length of 15 mm over 10 days after a latency period of 1 day. The distractors were then left in situ for 6–8 weeks for bony consolidation, after which they were removed via the same access incision.

A review of the medical records was undertaken to document demographic, operative, and perioperative data and also growth over the first postoperative year. The sample was divided into syndromic and nonsyndromic cohorts as designated by clinical genetics, and a descriptive analysis of the data was performed. Growth data were analyzed by calculating *Z*-scores of weight for corrected ages from the World Health Organization's published anthropometric data[23] at 5 time points: birth, distractor insertion and removal, and 6 and 12 months postoperatively. *Z*-scores of birth weight for preterm infants were calculated from the Fenton growth charts.[24] The means of these *Z*-scores were then calculated at each time point for the 2 cohorts to indicate their average growth. Failure to thrive was defined as being when weight dropped either 2 or more centile lines (a drop in *Z*-score of ≥1.34) or below the third centile line (a *Z*-score of <−2).[25] Where data were not normally distributed, they are presented as medians and interquartile ranges (IQRs).

For the second phase, all the patients were invited by letter to attend a clinical review and 39 families consented in writing to participate. A standardized assessment was undertaken by 2 researchers (A.N.A. and N.K.), which included a comprehensive dental examination with both facial and intraoral photographs. Teeth were charted as present or missing and classified as healthy, carious, or restored, and any anomalies such as ectopic position or developmental defects were noted. Radiographs were viewed only when already available. Where sufficient cooperation was possible, sensory deficits were explored with a cotton wool roll along the cutaneous distribution of the inferior alveolar nerve and the skin overlying the forehead and the body of the mandible posteriorly. The children were asked whether it felt normal, different, or "numb." These responses were mapped as normal sensation, hypoesthesia or paraesthesia, and anesthesia. Scar cosmesis was assessed using the Stony Brook Scar Evaluation Scale,[26] which includes 5 categories: width, height, color, suture marks, and overall appearance. Patients and/or their parents were also asked to make a subjective assessment of their scars on a Likert scale of 0 to 5, where 0 is unhappy.

RESULTS

Seventy-three children under the age of 5 years underwent MDO at the RCH between 2000 and 2012. Of these, 42 were designated as syndromic (Table 1). The oldest nonsyndromic child to undergo distraction was 32 months old, whereas the oldest syndromic child was 39 months old. All but one infant had congenital micrognathia. The exception was a child who developed micrognathia secondary to temporomandibular joint ankylosis, thought to be due to an early postnatal streptococcal septicemia. Two infants died at home after distraction therapy. One infant

Table 1. Demographics of Patients who Had MDO

	Nonsyndromic	Syndromic
Patients	31	42 10 Treacher Collins 6 stickler 4 Goldenhar 22 other
Age at MDO (mo)	2.0 (1.7–4.2)*	3.3 (2.1–7.4)*
<3	22	18
<12	3	17
<60	6	7

(Toriello-Carey syndrome), aspirated and suffered respiratory arrest during sleep before distractor removal and the other child (Treacher Collins syndrome), died 2 years postdistraction after a failed tracheostomy tube change.

Patients were extubated in the intensive care unit after a mean of 5.3±2.3 (SD) days and discharged home, 14.9±8.3 (SD) days after MDO. Two children had prolonged lengths of stay because of infection: one had a respiratory syncytial virus and the other a pseudomonas infection involving the distractor activation arms. Erythema of the skin surrounding the activation arm was reported in 30 patients (41%) but resolved quickly with conservative management. Four patients (5%) required early distractor removal because of infection at 6 weeks. Device failure occurred in 3 children (4%): 1 underwent reattachment of an activation arm and 2 others failed later in the distraction period. This occurred after resolution of airway obstruction, and no further intervention was required.

Of the children without tracheostomies, none required any further airway assistance (Fig. 1). Of the 8 tracheostomy-dependent infants before MDO, 5 (63%) were successfully decannulated within the first postoperative year, 1 was decannulated after 2 years, and 2 retained their tracheostomies. Seven (10%) patients, all syndromic, had undergone a second MDO procedure for further UAO. The shortest time to the second MDO procedure was 2 years with a median of 4.1 years (IQR, 2.2–6.8).

Sixty-one (83%) patients required supplemental feeding before MDO, with 55 (75%) requiring nasogastric tube (NGT) feeds. Six (8%) patients (all syndromic) had a percutaneous gastrostomy tube (PEG) (Fig. 2). The remaining children were fed orally with special care nursing bottles such as the Haberman. Forty-eight children (66%) were on an oral diet within 1-month post-MDO including 90% (28) of the nonsyndromic children. At the end of the first postoperative year, 56 patients (77%) were feeding orally, 5 (7%) had NGTs, and 12 (16%) had PEGs (including 7 who had NGTs before distraction but who then went on to have PEGs due to prolonged feeding difficulties).

The average growth of the patients followed within 1 centile line of their birth-predicted trajectories (Fig. 3). Complete data were available for 52 patients, and in this group, failure to thrive was observed in 5 (23%) of the nonsyndromic cohort and 8 (27%) of the syndromic cohort, before distraction. At 1-year post-MDO, growth had improved with 19 (86%) of the nonsyndromic cohort and 25 (83%) of the syndromic cohort growing within 2 centile lines of their weights at MDO. There was no statistical difference between the nonsyndromic and syndromic cohorts ($p > 0.05$), but the difference in growth velocities at distraction and 1-year post-MDO was statistically significant ($p = 0.006$), showing overall positive growth.

Of the 73 families invited for clinical review, 39 (53%) responded. The median age of the patients was 4.6 years (IQR, 3.2–9.5 y) for the nonsyndromic and 5.2 years (IQR, 3.5–10.1) for the syndromic groups, respectively (Table 2). Patients were at various stages of dental development with 22 (56%) in their primary dentition, 13 (33%) in their mixed dentition, and 4 (10%) in their permanent dentition. Twelve children (31%) had a healthy dentition with no signs of developmental defects of enamel (DDE). Twenty-five children (64%) had evidence of DDE and/or

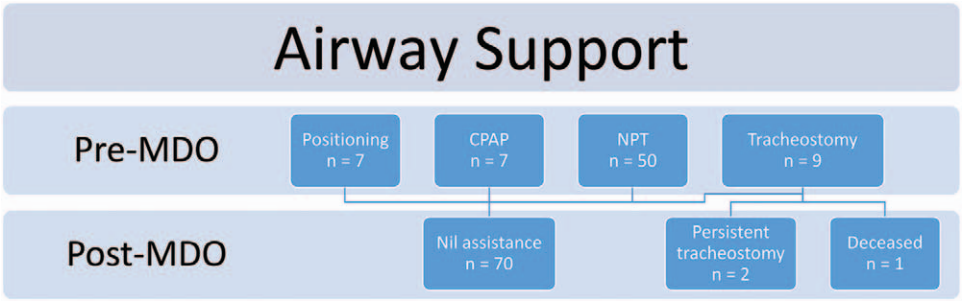

Fig. 1. Airway support requirements of the cohort before and after MDO. CPAP, continuous positive airway pressure; NPT, nasopharyngeal tube.

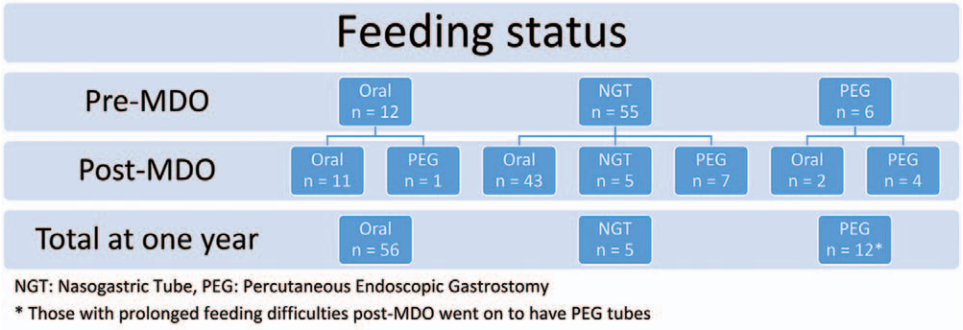

Fig. 2. Feeding status of the cohort pre-MDO and at 1-y post-MDO.

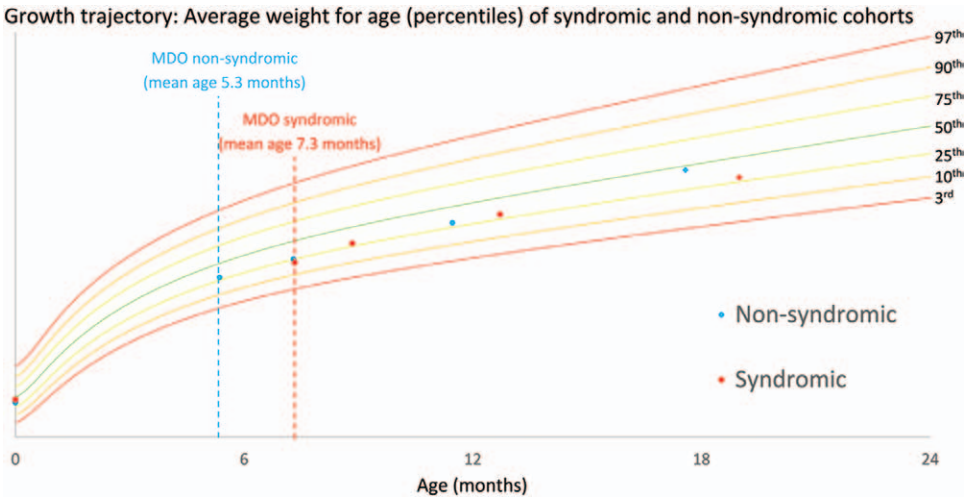

Fig. 3. Pediatric growth chart showing the average weight for age (percentiles) of syndromic and non-syndromic cohorts.

morphological changes in at least 1 tooth (Table 3). Of 12 patients who had hypomineralization defects in teeth proximal to the operative sites, 9 (75%) also had hypomineralization defects of teeth distant to the sites. Fourteen patients had hypoplastic defects of enamel in teeth near the operative site and 6 (43%) also had hypoplastic defects in other teeth. Twenty-seven patients (69%) were caries free and 2 patients (5%) had untreated dental caries. The remaining 11 patients (28%) had evidence of restorations and/or extractions.

The health of 52 mandibular second primary molars was recorded and 26 (50%) were healthy, 10 (19%) had hypomineralized opacities, and 8 (15%) had hypoplastic lesions. A further 7 (13%) second primary molars had been restored and 4 (8%) were recorded as missing (either extracted or congenitally absent). Of the 30 first permanent mandibular molars present and erupted, only 4 (13%) were healthy with the most common defect being enamel hypoplasia (Fig. 4). A further 3 (10%) mandibular first permanent molars had large restorations in situ and 8 (27%) had been extracted in a total of 5 patients. With 5 patients greater than 11 years at review, there were 6 erupted second permanent mandibular molars: 2 were sound, 3 had hypomineralization defects, and 1 was restored. A further 4 mandibular second permanent molars had been extracted because of various defects, one that appeared to be elongated (Fig. 5). Mandibular permanent second premolars seemed well preserved, with 6 of 8 erupted teeth being sound and 2 having hypomineralization defects.

Only 12 of the 39 patients were able to reliably participate in neurosensory testing, comparing light-touch sensation along the cutaneous distribution of the mental nerve to the supraorbital nerve. Five patients (13%) described hypoesthesia in some parts of the cutaneous distribution of the mental nerve, but only 1 patient was previously aware of it.

All patients and/or their parents rated their scars highly with a score of 3 or more on the Stony Brook Scar Evaluation Scale (Fig. 6). Scar cosmesis, as rated by the primary researcher (A.N.A.), was scored in the top half of the scale in 38 patients (97%). The most frequent negative comment regarding scars related to the width at the exit site of the activation arms. Hyperpigmentation of the scars occurred in 3 patients (8%) and also occurred at the activation arm exit wound.

Table 2. Demographics of Patients Who Returned for Clinical Review

	Nonsyndromic	Syndromic
Patients	21	18 2 Treacher Collins 3 Stickler 1 Goldenhar 12 other
Age at MDO (mo)	2.0 (1.7–4.2)*	3.0 (1.8–5.2)*
Age at review (y)	4.6 (3.2–9.5)*	5.2 (3.5–10.1)*

Data presented as counts or *median (interquartile range).

Table 3. Dental Data, per Patient, for Patients Who Returned for Clinical Review (n = 39)

	Nonsyndromic, n (%)	Syndromic, n (%)	Total, n (%)
Sound dentition (whole mouth)	5 (24)	7 (39)	12 (31)
Sound mandibular second primary molars	5 (24)	5 (28)	10 (26)
Sound mandibular first permanent molars	0 (0)	(0)	(0)
Developmental defects (whole mouth)	16 (76)	9 (50)	25 (64)
Developmental defects (all teeth near the osteotomy site*)	14 (67)	7 (39)	21 (54)
Developmental defects (mandibular first permanent molars)	7 (33)	3 (17)	10 (26)

*Teeth near the osteotomy site were considered to be the mandibular permanent second premolars, first molars, and second molars and primary second molars.

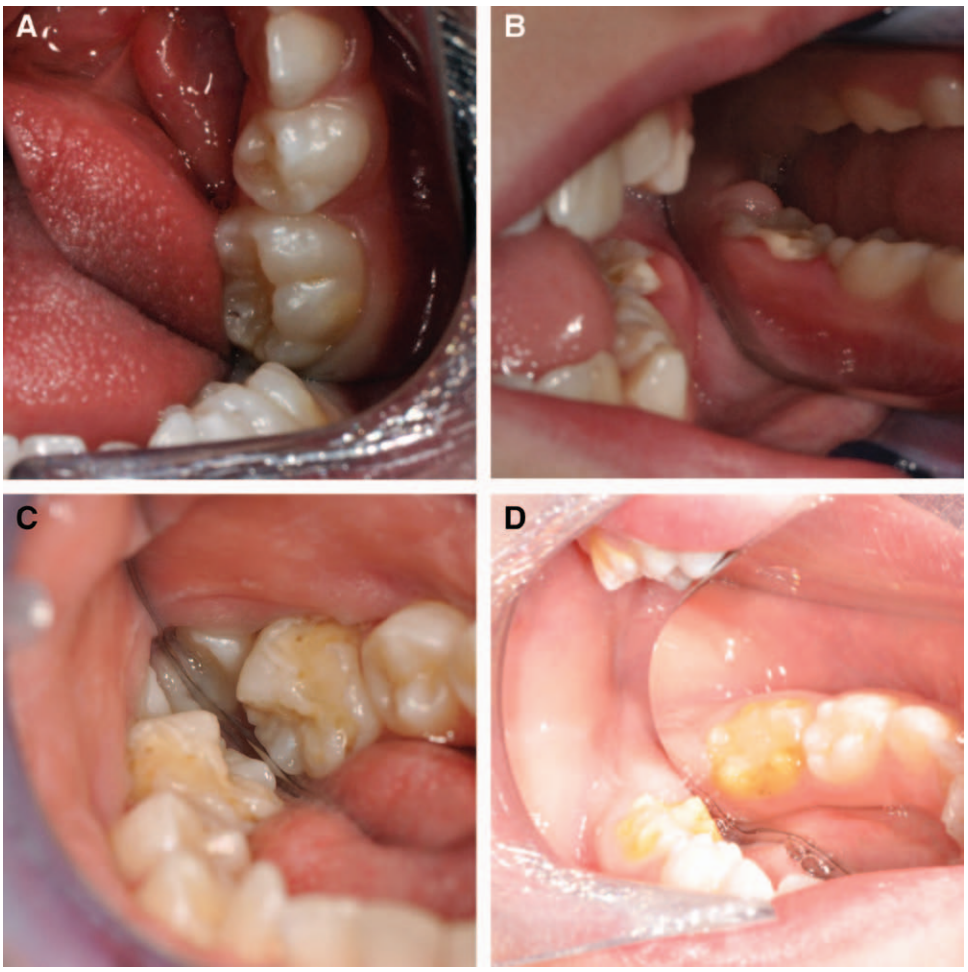

Fig. 4. Typical developmental defects of enamel observed in teeth near the operative site: buccal hypomineralization of lower left second primary molar (75) (A), buccal hypoplasia of lower left first permanent molar (36) (B), hypoplastic tooth lower right first permanent molar (46) (C) with abnormal morphology, and combined hypoplastic and hypomineralized defect of lower right first permanent molar (46) (D).

DISCUSSION

Traditionally, the management of UAO in micrognathic infants that failed to respond to nonsurgical therapy involved either a TLA or a tracheostomy.[27] MDO is a relatively new technique that has eliminated the need for tracheostomy in a large percentage of patients with a low complication rate, reduced inpatient stay, and decreased burden on families.[12,16,28–30] In this cohort, there was improvement in airway outcomes in the short term with 64 (87%) of the children (including all nonsyndromic children) being discharged from hospital postoperatively requiring no airway assistance. Among the 9 children who were tracheostomy-dependent before MDO, 6 were decannulated (5 within 1 y of MDO). The average total time from tracheostomy to decannulation of 29 months (range, 16 – 45 mo) compares favorably with a previous study in a center where MDO was unavailable in which the average total tracheostomy time was 37 months (range, 14 – 60 mo).[31] In a recent comparison of tracheostomy rates arising from 2 different protocols for management of UAO, a quarter (27%) of infants who underwent TLA went on to tracheostomy compared with none who underwent MDO.[18] These figures are similar to those reported by Lam et al[32] in which 76% of their cohort either avoided a tracheostomy completely or were decannulated postoperatively. Some patients in this study underwent repeated episodes of MDO for resolution of their UAO. This was also reflected in our cohort with 7 patients (10%) having a second MDO procedure, all of whom were syndromic.

Feeding difficulties are common among micrognathic infants and are related to the severity of the UAO.[33] Understanding the impact of MDO on feeding is limited with some suggesting that feeding problems persist postoperatively,[34] whereas others report improved feeding.[35–37] The feeding outcomes in this study were overwhelmingly positive, with 77% of the children changing to an all-oral diet within 1 year. Only 1, nonsyndromic, child continued supplemental feeding (NGT) at 1 year but changed to an all-oral diet within the next 4 months.

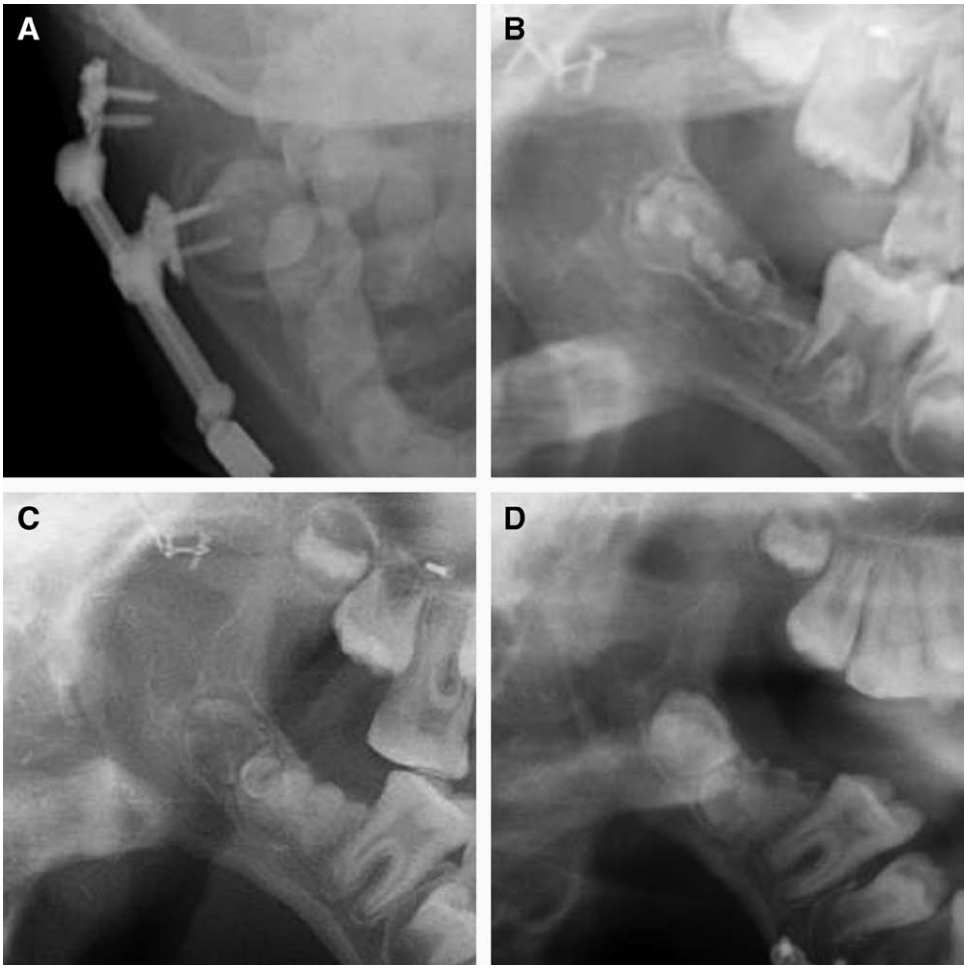

Fig. 5. Example of a local dental anomaly associated with MDO: patient at 3 y with the distractor in situ (A), the same patient at 6 y with what appears to be an elongated second permanent molar tooth bud (B), again at 9 y with the third permanent molar tooth bud developing (C), and finally at 12 y with apparently normal third molar development but abnormal appearance to the second permanent molar, which was subsequently removed (D). The first permanent molar exhibited some distobuccal hypoplasia but was in function, unrestored.

Few studies have addressed the impact of MDO on growth outcomes in this patient population. However, it has been shown that rates of failure to thrive are also related to the severity of UAO, improving with nasopharyngeal tube intubation[38,39] and TLA.[40] In 1 study of ten syndromic infants, MDO seemed to have limited impact on growth,[34] but the presence of comorbidities and the intrinsic growth limitation in those with syndromic diagnoses compromise these findings. In this cohort, failure to thrive was noted in thirteen (25%) infants before distraction, and this number decreased to 8 (15%), 12 months post MDO. However, as many of these infants were supplemented before MDO in the neonatal unit, specifically to optimize weight gain, these data need to be interpreted with caution.

High rates of dental anomalies, such as destruction or displacement of tooth follicles, have been previously reported in the literature.[20] The RCH surgical protocol specifically aims to minimize the risk of damage to the developing dentition. Timing of the surgical intervention is likely to impact on the nature of the damage to the teeth; the earlier the intervention in relation to tooth development the more likely the affected teeth will show a quantitative (or hypoplastic) defect (Fig. 4C) as opposed to a qualitative (or hypomineralized) opacity (Fig. 4A). The median age at the time of MDO for this cohort was between 2 and 3 months (Table 1) at which time the first permanent molars are very immature and in the early secretory phase of enamel formation and the defects are mainly hypoplastic in nature.[41,42] The development of second primary molars at this age is more advanced and their enamel is undergoing maturation. Consequently, the defects in these teeth were more commonly hypomineralized in nature.[41,42] One limitation of this study is that over half of the patients (56%) were still in their primary dentition at the time of clinical assessment, many without radiographs, making it impossible to assess congenitally absent or unerupted grossly displaced teeth in these patients. However, of those teeth that had erupted, 48% of the second prima-

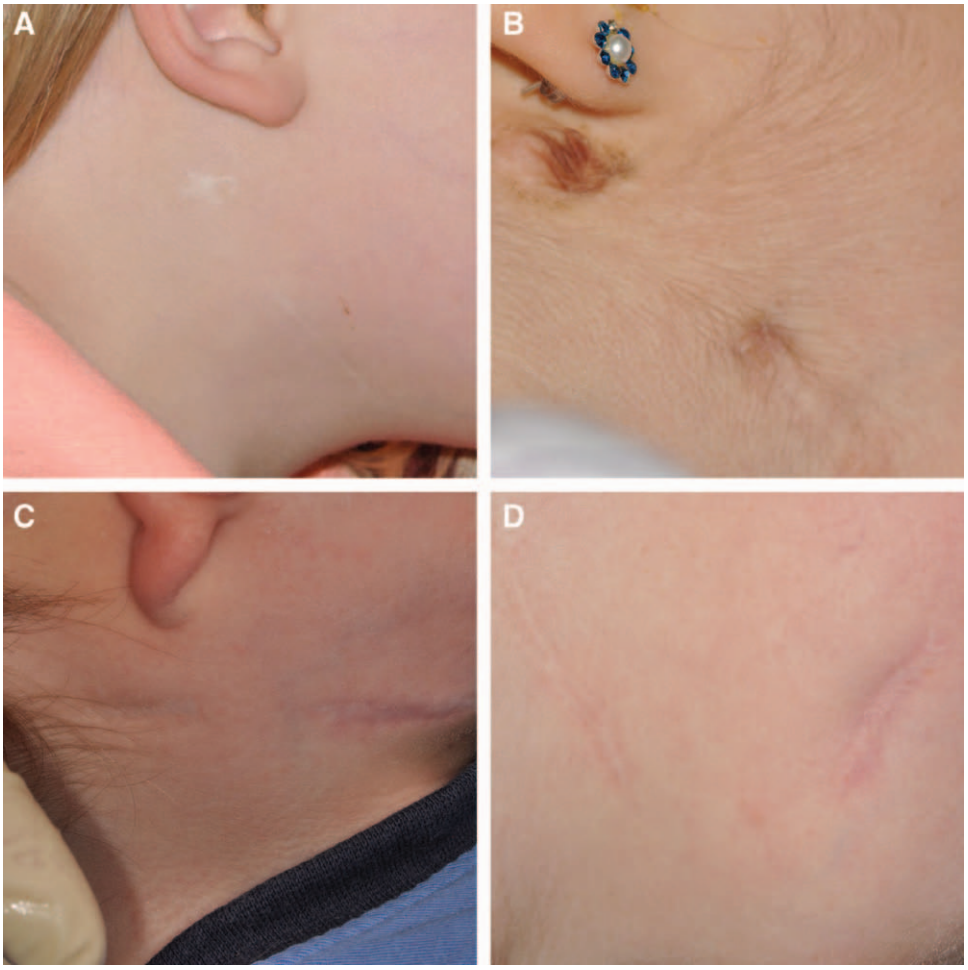

Fig. 6. Scar cosmesis: typical appearance of scars post-MDO (Stony Brook score, 5) (A), hyperpigmentation and increased width of scars (score, 3) (B), unesthetic scar with hyperpigmentation and increased width (score, 2) (C), and scar depression (score, 4) (D).

ry molars and 83% of the first permanent molars were either restored or had been extracted, suggesting that these teeth were also significantly compromised. DDE, most commonly hypomineralized defects, were also noted in teeth distant to the operative site. This suggests that hypomineralized defects may have a greater association with other systemic factors, whereas enamel hypoplasia may be the result of localized insult such as surgical trauma. Certainly, there is growing recognition of the impact of systemic insults, such as preterm birth, low birth weight, and perinatal illnesses on tooth development,[43] all of which are putatively more common in this cohort of children.[44]

Scar cosmesis was considered acceptable by all participants. This is in contrast to an earlier study in which hypertrophic scarring occurred in 0.67% to 15.6% of patients.[45] Differences in surgical protocol may account for this, with the RCH protocol involving internal rather than external distractors. Carers further reported improved cosmesis with continued growth, as the submandibular scars migrated inferiorly further reducing in visibility (Fig. 7).

Inferior alveolar nerve injury after infant MDO has not been well described in the literature. In a questionnaire study, inferior alveolar nerve hypoesthesia was reported in 19.5% of patients where distraction rates were greater than 1 mm per day, which dropped to 2.4% when distraction was 1 mm per day or slower.[46] In this study, hypoesthesia was present in 5 patients (42% of those able to describe their sensation), although only 1 was previously aware of it.

CONCLUSIONS

Mandibular distraction seems to be a predictable and effective technique for managing micrognathia when associated with UAO. All nonsyndromic Robin sequence patients were discharged within 2 weeks without further need for airway support. However, although initially successful, second distraction procedures were necessary in a proportion of the syndromic children where both their anatomy and medical comorbidities made resolution of UAO more demanding. There was a low rate of damage to the teeth that could be directly attributed to the MDO procedure. However, as the majority of patients were still in the primary dentition, on-

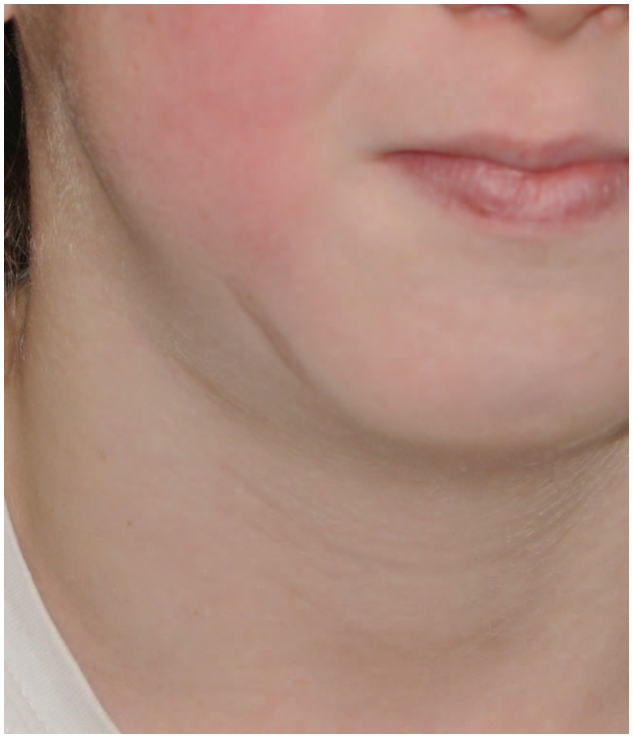

Fig. 7. Scar migration where labiomental and submandibular scars migrated to the neck becoming less visible from the front (score 4 due to increased width of anterior scar).

going dental review remains important. Parents should be warned about the potential longer-term implications of MDO and the fact that further treatment for airway management may be required. Further prospective studies are important to establish an evidence base to the management of this population.

Nicky Kilpatrick, BDS, PhD, FRACDS(Paeds)
Murdoch Childrens Research Institute
Royal Children's Hospital Melbourne
Flemington Rd
Parkville, Victoria 3052, Australia.
E-mail: nicky@bassdata.com.au

ACKNOWLEDGMENTS

We thank Supriya Raj, Lochana Ramalingam, Susie Gibb, and Suzanne Glare for their invaluable assistance and support with the research process and clinical review. Medical illustrations are provided courtesy of the Education Resource Centre of the Royal Children's Hospital.

REFERENCES

1. Online Mendelian Inheritance in Man, OMIM®, [Internet]. MD: McKusick-Nathans Institute of Genetic Medicine, Johns Hopkins University. Available at: http://omim.org/. Accessed February 1, 2015.
2. Robin P. A fall of the base of the tongue considered as a new cause of nasopharyngeal respiratory impairment: Pierre Robin sequence, a translation. 1923. *Plast Reconstr Surg.* 1994;93:1301–1303.
3. Tan TY, Kilpatrick N, Farlie PG. Developmental and genetic perspectives on Pierre Robin sequence. *Am J Med Genet C Semin Med Genet.* 2013;163C:295–305.
4. Evans KN, Sie KC, Hopper RA, et al. Robin sequence: from diagnosis to development of an effective management plan. *Pediatrics* 2011;127:936–948.
5. Abel F, Bajaj Y, Wyatt M, et al. The successful use of the nasopharyngeal airway in Pierre Robin sequence: an 11-year experience. *Arch Dis Child* 2012;97:331–334.
6. Anderson KD, Cole A, Chuo CB, et al. Home management of upper airway obstruction in Pierre Robin sequence using a nasopharyngeal airway. *Cleft Palate Craniofac J.* 2007;44:269–273.
7. Olson TS, Kearns DB, Pransky SM, et al. Early home management of patients with Pierre Robin sequence. *Int J Pediatr Otorhinolaryngol.* 1990;20:45–49.
8. Daniel M, Bailey S, Walker K, et al. Airway, feeding and growth in infants with Robin sequence and sleep apnoea. *Int J Pediatr Otorhinolaryngol.* 2013;77:499–503.
9. Waters KA, Everett F, Bruderer J, et al. The use of nasal CPAP in children. *Pediatr Pulmonol Suppl.* 1995;11:91–93.
10. Gerzanic L, Feichtinger M, Kärcher H. The influence of the Tübingen soft palate plate and early cleft closure on the nasopharyngeal airway for the management of airway obstruction in an infant with Pierre Robin sequence: A case report. *Int J Surg Case Rep.* 2012;3:608–610.
11. Kirschner RE, Low DW, Randall P, et al. Surgical airway management in Pierre Robin sequence: is there a role for tongue-lip adhesion? *Cleft Palate Craniofac J.* 2003;40:13–18.
12. Genecov DG, Barcelo CR, Steinberg D, Trone T, Sperry E. Clinical experience with the application of distraction osteogenesis for airway obstruction. *J Craniofac Surg.* 2009;20(Suppl 2):1817–1821.
13. Denny AD, Amm CA, Schaefer RB. Outcomes of tongue-lip adhesion for neonatal respiratory distress caused by Pierre Robin sequence. *J Craniofac Surg.* 2004;15:819–823.
14. Hopkins C, Whetstone S, Foster T, et al. The impact of paediatric tracheostomy on both patient and parent. *Int J Pediatr Otorhinolaryngol.* 2009;73:15–20.
15. Pereira KD, MacGregor AR, Mitchell RB. Complications of neonatal tracheostomy: a 5-year review. *Otolaryngol Head Neck Surg.* 2004;131:810–813.
16. Denny A, Kalantarian B. Mandibular distraction in neonates: a strategy to avoid tracheostomy. *Plast Reconstr Surg.* 2002;109:896–904; discussion 905.
17. Denny AD, Talisman R, Hanson PR, Recinos RF. Mandibular distraction osteogenesis in very young patients to correct airway obstruction. *Plast Reconstr Surg.* 2001;108:302–311.
18. Flores RL, Tholpady SS, Sati S, et al. The surgical correction of Pierre Robin sequence: mandibular distraction osteogenesis versus tongue-lip adhesion. *Plast Reconstr Surg.* 2014;133:1433–1439.
19. Paes EC, Mink van der Molen AB, Muradin MS, et al. A systematic review on the outcome of mandibular distraction osteogenesis in infants suffering Robin sequence. *Clin Oral Investig.* 2013;17:1807–1820.
20. Kleine-Hakala M, Hukki J, Hurmerinta K. Effect of mandibular distraction osteogenesis on developing molars. *Orthod Craniofac Res.* 2007;10:196–202.
21. Hong P, Graham E, Belyea J, et al. The long-term effects of mandibular distraction osteogenesis on developing deciduous molar teeth. *Plast Surg Int.* 2012;2012:913807.
22. Chigurupati R, Massie J, Dargaville P, et al. Internal mandibular distraction to relieve airway obstruction in infants and young children with micrognathia. *Pediatr Pulmonol.* 2004;37:230–235.
23. de Onis M, Onyango AW, Borghi E, et al. Development of a WHO growth reference for school-aged children and adolescents. *Bull World Health Organ* 2007;85:660–667.
24. Fenton TR, Kim JH. A systematic review and meta-analysis to revise the Fenton growth chart for preterm infants. *BMC Pediatr.* 2013;13:59.

25. Wright CM. Identification and management of failure to thrive: a community perspective. *Arch Dis Child* 2000;82:5–9.
26. Singer AJ, Arora B, Dagum A, et al. Development and validation of a novel scar evaluation scale. *Plast Reconstr Surg.* 2007;120:1892–7.
27. Collins B, Powitzky R, Robledo C, et al. Airway management in pierre robin sequence: patterns of practice. *Cleft Palate Craniofac J.* 2014;51:283–289.
28. Ow AT, Cheung LK. Meta-analysis of mandibular distraction osteogenesis: clinical applications and functional outcomes. *Plast Reconstr Surg.* 2008;121:54e–69e.
29. Paes EC, Fouché JJ, Muradin MS, et al. Tracheostomy versus mandibular distraction osteogenesis in infants with Robin sequence: a comparative cost analysis. *Br J Oral Maxillofac Surg.* 2014;52:223–229.
30. Tahiri Y, Viezel-Mathieu A, Aldekhayel S, et al. The effectiveness of mandibular distraction in improving airway obstruction in the pediatric population. *Plast Reconstr Surg.* 2014;133:352e–359e.
31. Tomaski SM, Zalzal GH, Saal HM. Airway obstruction in the Pierre Robin sequence. *Laryngoscope* 1995;105:111–114.
32. Lam DJ, Tabangin ME, Shikary TA, et al. Outcomes of mandibular distraction osteogenesis in the treatment of severe micrognathia. *JAMA Otolaryngol Head Neck Surg.* 2014;140:338–345.
33. Li HY, Lo LJ, Chen KS, et al. Robin sequence: review of treatment modalities for airway obstruction in 110 cases. *Int J Pediatr Otorhinolaryngol.* 2002;65:45–51.
34. Spring MA, Mount DL. Pediatric feeding disorder and growth decline following mandibular distraction osteogenesis. *Plast Reconstr Surg.* 2006;118:476–482.
35. Hong P, Brake MK, Cavanagh JP, et al. Feeding and mandibular distraction osteogenesis in children with Pierre Robin sequence: a case series of functional outcomes. *Int J Pediatr Otorhinolaryngol.* 2012;76:414–418.
36. Monasterio FO, Molina F, Berlanga F, et al. Swallowing disorders in Pierre Robin sequence: its correction by distraction. *J Craniofac Surg.* 2004;15:934–941.
37. Papoff P, Guelfi G, Cicchetti R, et al. Outcomes after tongue-lip adhesion or mandibular distraction osteogenesis in infants with Pierre Robin sequence and severe airway obstruction. *Int J Oral Maxillofac Surg.* 2013;42:1418–1423.
38. Heaf DP, Helms PJ, Dinwiddie R, et al. Nasopharyngeal airways in Pierre Robin Syndrome. *J Pediatr.* 1982;100:698–703.
39. Pandya AN, Boorman JG. Failure to thrive in babies with cleft lip and palate. *Br J Plast Surg.* 2001;54:471–475.
40. Cozzi F, Totonelli G, Frediani S, et al. The effect of glossopexy on weight velocity in infants with Pierre Robin syndrome. *J Pediatr Surg.* 2008;43:296–298.
41. Nanci A, Ten Cate A. *Ten Cate's Oral Histology: Development, Structure, and Function.* 6th ed. St. Louis: Mosby; 2003.
42. Suckling GW. Developmental defects of enamel–historical and present-day perspectives of their pathogenesis. *Adv Dent Res.* 1989;3:87–94.
43. Seow WK. Developmental defects of enamel and dentine: challenges for basic science research and clinical management. *Aust Dent J.* 2014;59(Suppl 1):143–154.
44. Crombie F, Manton D, Kilpatrick N. Aetiology of molar-incisor hypomineralization: a critical review. *Int J Paediatr Dent.* 2009;19:73–83.
45. Master DL, Hanson PR, Gosain AK. Complications of mandibular distraction osteogenesis. *J Craniofac Surg.* 2010;21:1565–1570.
46. Mofid MM, Manson PN, Robertson BC, et al. Craniofacial distraction osteogenesis: a review of 3278 cases. *Plast Reconstr Surg.* 2001;108:1103–1115.