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An unusual complication of mandibular distraction

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Summary. *Background.* The authors present an unusual complication of mandibular distraction in a child with the curious condition of multiple pterygium syndrome is presented.
Case report. The patient was a Caucasian male with severe pterygia in his neck. As a result of his limited mouth opening and restricted upper airway leading to obstruction, he underwent lengthening of his mandible by distraction, which significantly improved his breathing. During his follow-up, it was observed that an unusually elongated permanent molar was present in an abnormal position.
Conclusion. This case highlights the need to carefully plan the sites for osteotomy and the potential for damage to the developing permanent dentition in young children.

Introduction

Multiple pterygium syndrome is comprised of severe neck contracture and other craniofacial anomalies. The associated mandibular hypoplasia may exacerbate the upper airway obstruction. Mandibular distraction is an established method to improve the upper airway in young children with airway obstruction caused by mandibular hypoplasias, such as Pierre Robin sequence, Treacher Collins syndrome and Nager syndrome [1–3]. Mandibular distraction has been shown to increase the minimum cross-sectional area of the upper airway that is maintained throughout the period of growth [1]. However, mandibular distraction can also be associated with complications including infection, haemorrhage, incomplete osteotomy, dislodgement of pins, failure of distraction, granuloma, abscess and inferior alveolar nerve damage, and requires great care in young children [4].

The authors report what they believe to be a previously unreported complication following mandibular distraction in a 3-year-old boy with multiple pterygium syndrome and Pierre Robin sequence. They present a long-term complication of distortion of a permanent tooth follicle.

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Case report

A 3-year-old boy presented to the Australian Craniofacial Unit, Women's and Children's Hospital, North Adelaide, South Australia, Australia, with multiple pterygium syndrome, Pierre Robin Sequence and cleft secondary palate. The subject had severe contractures in his neck, axillae, elbows, wrists, groins, knees and ankles. Other facial findings included a scaphocephalic head, bilateral epicanthal folds, antimongoloid slant and malposition of the ears (Fig. 1). He had a hypoplastic lower jaw with his chin almost adherent to the sternum, and obstructive sleep apnoea as a result of a small nasopharyngeal passage.

Oral examination was difficult because of the limited mouth opening. The patient had previously required multiple dental extractions, which were carried out for dental caries that was a result of poor hygiene exacerbated by limited mouth opening. He underwent multidisciplinary assessment, including a review by a respiratory physician. A formal sleep study reported significant episodes of obstructive apnoea. Plain radiographs, a computed tomography scan, three-dimensional reconstruction and magnetic resonance imaging to assist in evaluating his underlying anatomical structures supplemented his investigations.

The subject subsequently underwent bilateral mandibular osteotomy, and distraction was performed at the rate of one millimetre per day. A mandibular lengthening of 12 mm was achieved (Figs 2 and 3). After stabilization for 3 weeks, the distracters were

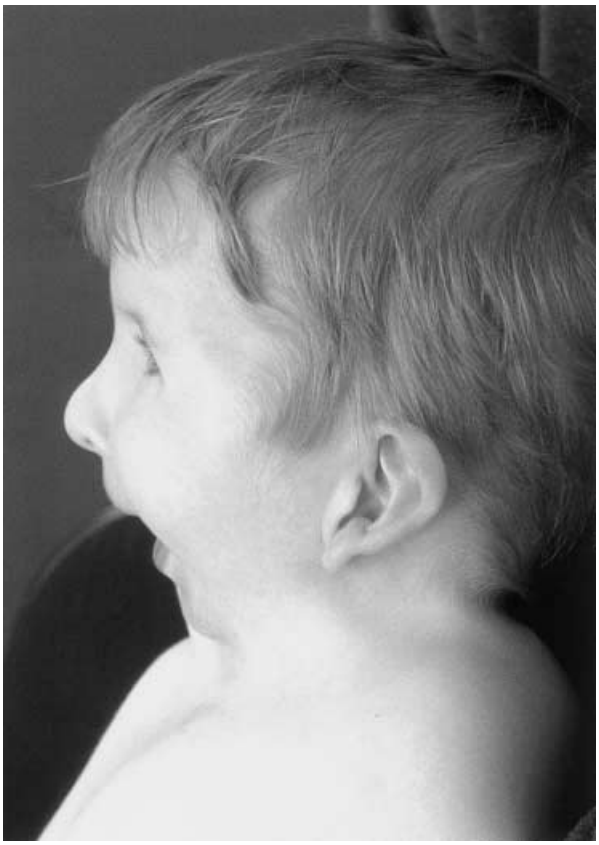

Fig. 1. Photograph of the subject showing severe pterygia of the neck and a hypoplastic mandible.

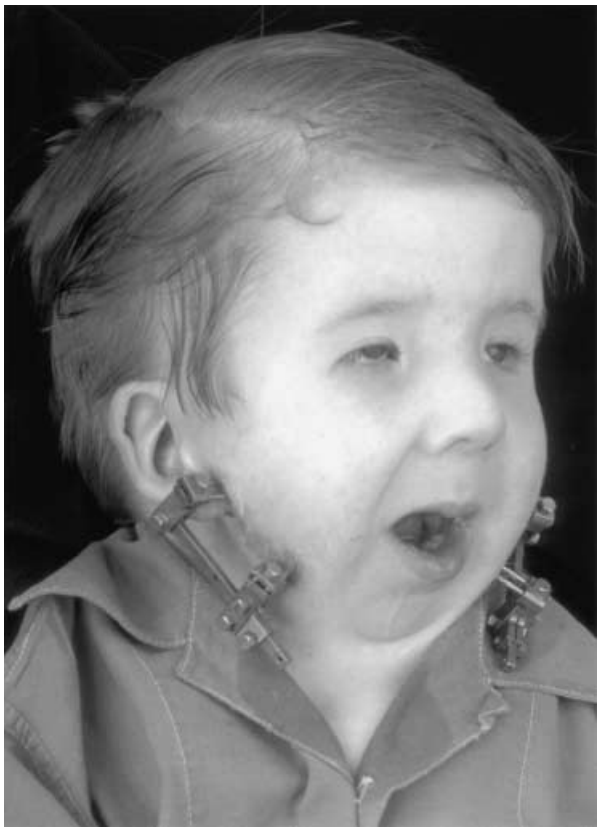

Fig. 2. Photograph showing limited mouth opening.

removed and his cleft palate was repaired at the same time. A subsequent sleep study indicated no further apnoeic episodes. The patient was also noted to have improved mouth opening and improved feeding. He has been kept under regular review, and his airway has remained patent.

However, an orthopantomogram at the age of 8 years revealed a curious elongated permanent molar lying in an abnormal position that was symptomless (Fig. 4). A review of the radiograph demonstrated that the first permanent molar was involved in the osteotomy site and was itself distracted. All radiology films were retrospectively reviewed, and it was noted that he had a well-developed crown of left lower molar prior to osteotomy, as shown in Fig. 5. Subsequent radiological examination revealed that the osteotomy site did not involve the crown of the molar on the left side (Fig. 6). His remaining dentition had unremarkable dental anatomy.

The subject's mouth opening deteriorated, and as a part of his subsequent management at the 11 years of age, a nylon model was constructed to plan his surgery. This revealed grossly hypertrophied coronoid processes, a markedly hypoplastic mandible and a very obtuse body ramus angle (Fig. 7).

The patient subsequently underwent stage release of his neck contracture using skin graft, which permitted improvement of his mouth opening. He will require further release of his neck contracture and will be continuously reviewed by the multidisciplinary team according to the Craniofacial protocol throughout the growth period.

Discussion

Mandibular distraction is an established procedure to improve the upper airway in Pierre Robin sequence, Treacher Collins syndrome, Nager syndrome and other similar conditions with apnoeic attacks [1–3]. The presence of severe obstructive apnoea, which was identified in this patient by the respiratory physician, with pterygium syndrome directed the authors to lengthen the mandible by distraction instead of releasing the neck contracture as a first stage to facilitate mandibular growth potential.

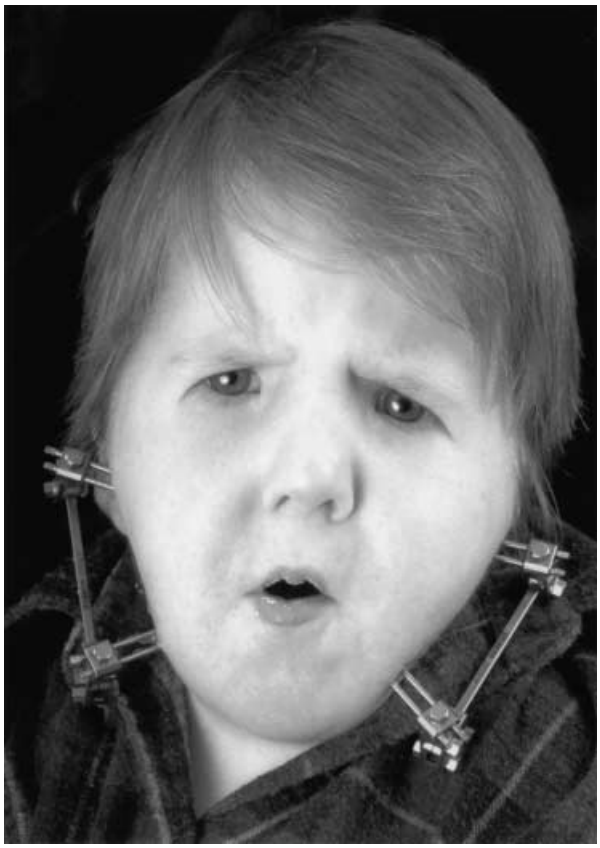

Fig. 3. Photograph showing linear mandibular distraction.

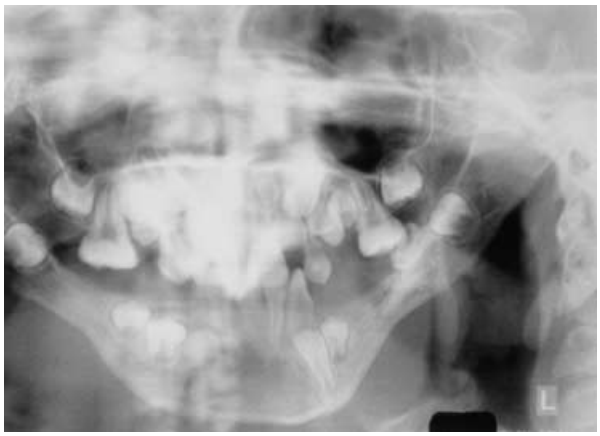

Fig. 4. Orthopantograph showing the elongated permanent molar on the left side.

Multiple pterygium syndrome is a very rare congenital deformity with autosomal recessive inheritance manifested by multiple flexural contractures and secondary deformities involving the craniofacial region, spine, trunk, anogenital region and limbs [5]. The causes are probably heterogeneous, but decreased foetal movement and neuromuscular pathology have been suggested [6].

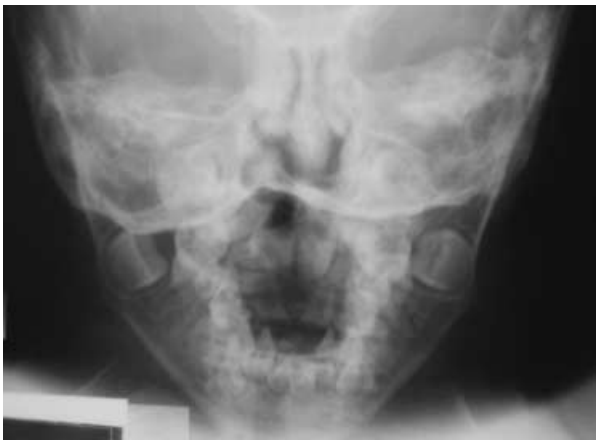

Fig. 5. Plain X-ray of the mandible prior to the osteotomy done when the subject was 3 years of age showing the well-developed crown of the left lower molar.

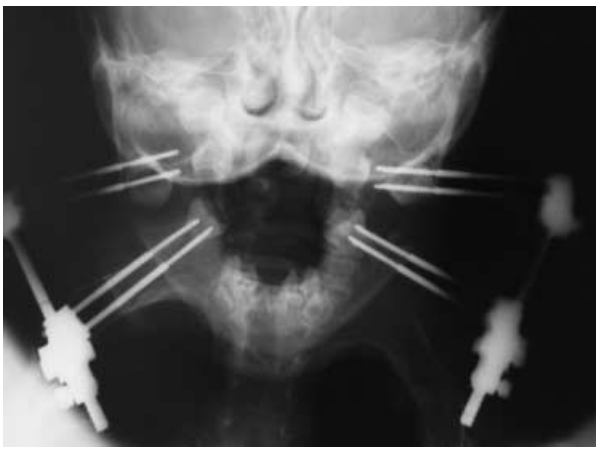

Fig. 6. Plain X-ray of the mandible after the osteotomy showing the intact lower molar on the left side.

Distraction of tissues has been utilized to recruit new bone and soft tissues following the principle of Ilizarov. Tissues including bone, muscle and tendon have been reported to undergo histogenesis during the distraction [7]. In this case, it would appear that the distraction force not only stretched these tissues, but may have elongated Hertwig's root sheath of the permanent molar as well. The dental development at the age of 3 years in his case was within normal limits. The other curious finding of the nylon model of this patient was the grossly hypertrophied coronoid process. It indicates that the temporalis muscle generated large forces over a prolonged period

Fig. 7. Nylon model made when the subject was 11 years of age showing the hypertrophied coronoid process.

to achieve mouth opening, resulting in such an anatomical change (Fig. 5).

In summary, the authors report a late complication following mandibular distraction resulting in damage to the developing dentition, and it highlights the need for careful positioning of the osteotomy and the placement of pins. Careful review of dental radiographs should be undertaken to establish the position of the dental follicle as part of the preoperative planning of the osteotomy site.

What this paper adds

- This paper presents a previously unreported long-term complication of mandibular distraction. Distraction as a part of airway management in those with mandibular hypoplasia may inevitably result in distortion of developing tooth buds.

Why this paper is important for paediatric dentists

- Paediatric dentists are an integral part of the multidisicplinary team that manages patients undergoing mandibular distraction. Mandibular distraction may inevitably alter the dental root morphology which has clinical implication should extraction be required.

References

1 Anderson PJ, Netherway DJ, Abbott A, Moore M, David DJ. Mandibular lengthening by distraction for airway obstruction in Treacher-Collins syndrome: the long-term results. *Journal of Craniofacial Surgery* 2004; **15**: 47–50.

2 Moore MH, Guzman-Stein G, Proudman TW, Abbott AH, Netherway DJ, David DJ. Mandibular lengthening by distraction for airway obstruction in Treacher-Collins syndrome. *Journal of Craniofacial Surgery* 1994; **5**: 22–25.

3 Denny AD, Talisman R, Hanson PR, Recinos RF. Mandibular distraction osteogenesis in very young patients to correct airway obstruction. *Plastic and Reconstructive Surgery* 2001; **108**: 302–311.

4 Davies J, Turner S, Sandy JR. Distraction osteogenesis – a review. *British Dental Journal* 1998; **185**: 462–467.

5 Soranger S, Spranger M, Meinck HM, Tariverdian G. Two sisters with Escobar syndrome. *American Journal of Medical Genetics* 1995; **57**: 425–428.

6 Ozkinay FF, Ozkinay C, Akin H, Azarsiz S, Gunduz C. Multiple pterygium syndrome. *Indian Journal of Paediatrics* 1997; **64**: 113–116.

7 Murray JH, Fitch RD. Distraction histogenesis: principles and indications. *Journal of the American Academy of Orthopaedic Surgeons* 1996; **4**: 317–327.