CASE REPORT

Correction of Airway Obstruction in Congenital Micrognathia by Mandibular Distraction Osteogenesis

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ABSTRACT

Varying degrees of upper airway obstruction is almost universally present in patients of congenital micrognathia which needs to be corrected as early as possible. This allows appropriate feeding and growth and prevents long-term complications such as pulmonary hypertension and cor pulmonale. We report the case of a tracheostomy-dependent, 4-year-old child with congenital micrognathia who was treated with mandibular distraction osteogenesis. This is the treatment of choice for surgical correction of mandibular hypoplasia and for the challenging airway management in infants. Once a bone length of 2 cm was achieved through distraction osteogenesis, the child was completely relieved of respiratory obstruction and tracheostomy tube was removed through the process of decannulation.

Key words: *Congenital micrognathia. Respiratory obstruction. Tracheostomy. Distraction osteogenesis. Mandible.*

INTRODUCTION

Congenital micrognathia as a cause for airway obstruction is a very challenging problem encountered in neonate with craniofacial disorders. There are various syndromic and non-syndromic causes for micrognathia in the neonates. Syndromes frequently presenting with upper airway obstruction include the Pierre Robin syndrome, Treacher Collins syndrome, Crouzon syndrome, Nager syndrome, and velocardiofacial syndrome. Clinical signs and symptoms of obstruction may include positional inspiratory obstruction, episodic apnea, decreased oxygen saturation, carbon dioxide retention, insufficient feeding or complete obstruction, leading to respiratory arrest. Early recognition and treatment of obstruction is necessary to allow appropriate feeding and growth, and to prevent long term problems such as pulmonary hypertension and cor pulmonale.[1]

Various surgical and non-surgical treatment methods have been advised for the treatment of this airway obstruction.[2] Mandibular distraction osteogenesis (MDO) is an orthopaedic technique of new bone formation by gradual traction of a fracture callus formed between osteotomized bony segments. It is becoming a treatment of choice for the surgical correction of facial asymmetry, micrognathia, mandibular hypoplasia of different etiology,[3] and thus for the challenging airway management in infants.[2-4]

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Received March 19, 2010; accepted May 12, 2011.

Its contribution to early decannulation of tracheostomy-dependent patients has been well documented.[5] Orthodontics plays a pivotal role in correction of post-distraction occlusal changes.[6]

We report a case of a tracheostomy-dependent 4-year-old child with congenital micrognathia, treated with bilateral MDO, at the Armed Forces Institute of Dentistry, Rawalpindi, Pakistan.

CASE REPORT

A 4-year-old male child was referred at the Department of Oral and Maxillofacial Surgery, Armed Forces Institute of Dentistry, Rawalpindi, in August 2009. The patient had undergone tracheostomy at the age of 18 months due to breathing difficulties, blocked nasal airway, recurrent respiratory infections and persistent stridor since birth. The physical development of the child was far behind his age (Figure 1). After thorough examination by maxillofacial surgery team and considering that the child's main problem was the micro- and retrognathia, (which led to the placement of the

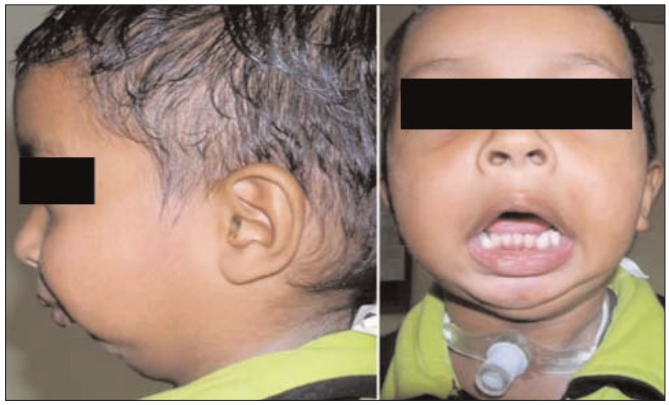

Figure 1: A 4-year-old micrognathic child who has undergone tracheostomy to relieve persistent stridor and upper respiratory obstruction.

floor of the mouth and tongue posteriorly), a therapeutic augmentation of the mandible via distraction osteogenesis was planned. For this purpose an internal unidirectional, titanium, distractor device of treu dynamic (Germany) brand was chosen. Radiological evaluation to help plan the operation, included orthopantomogram (Figure 2), lateral and anterior-posterior cephalograms, as well as CT scan. It was decided that the internal distractor would be placed bilaterally in the posterior mandibular body close to the angle, through an intra oral approach.

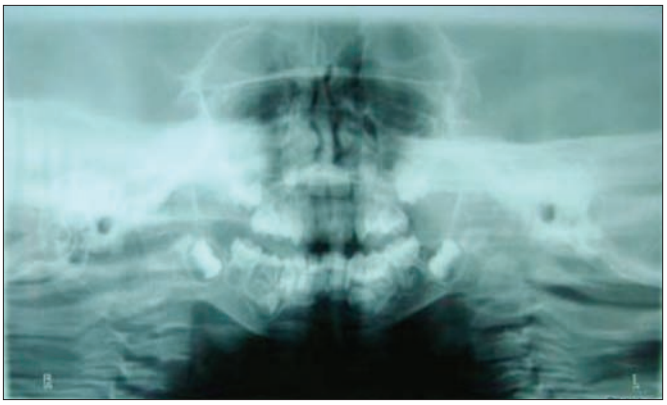

Figure 2: Pre-operative (i.e. pre-distraction) orthopantomogram of the patient.

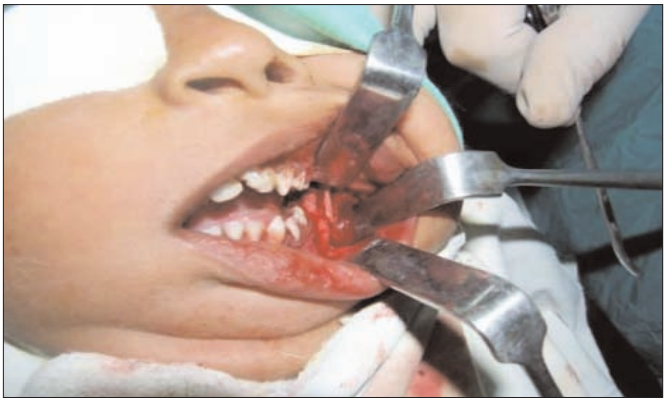

Figure 3: Operative site exposed through intraoral approach for positioning of distractor device.

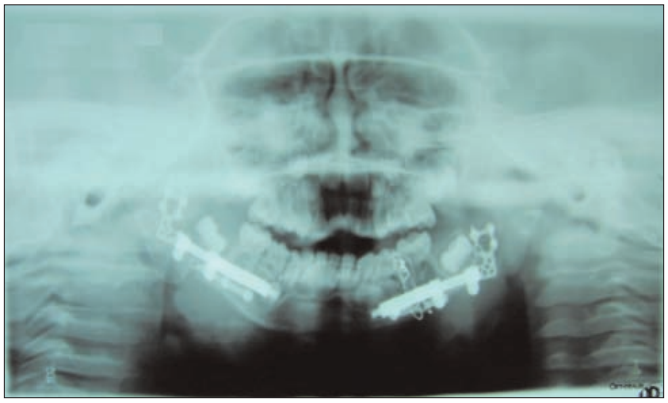

Figure 4: Orthopantomogram showing distractor devices in place bilaterally and a gain of 2 cm bone length.

The child was operated under general anaesthesia through the tracheostomy. Under aseptic conditions, the operative site was approached through vestibular incision extending up to anterior border of ramus, the distractor device was positioned, screws were inserted prior to osteotomy. The device was removed and osteotomy was performed (Figure 3).

Then the distractor was fixed and its function was checked. The incision was closed by careful suturing with 3/0 vicryl. The same procedure was performed on the contralateral side.

After waiting for a 3 days latency period, active distraction at a rate of 1 mm/day at each side commenced on the 4th postoperative day and lasted for 20 days. The parents were instructed regarding activation and handling of the device. After a consolidation period of 4 weeks the patient was examined clinically and radiologically to confirm ossification of the distraction sites and a bilateral length gain of 2 cm (Figure 4).

As the mandible increased in size and the tongue was allowed more space anteriorly, normal breathing became possible. The tracheostomy tube was removed 3 weeks postoperatively through the process of decannulation after consultation with ENT department of Combind Military Hospital, Rawalpindi. The distractors were removed 2 months postoperatively under general anaesthesia. Monthly follow-up will continue for next 2 years.

DISCUSSION

Varying degrees of upper airway obstruction is almost universally present in patients of congenital micrognathia which needs to be corrected as early as possible to prevent long-term problems such as pulmonary hypertension and cor pulmonale.[1-5]

Various options for airway management are available, aiming at moving the tongue base anteriorly out of the airway. The most conservative measure is placing the child in prone position to help displace the tongue anteriorly out of the airway and the use of nasopharyngeal airways. Among the available surgical options; there are soft tissue and skeletal procedures that relieve respiratory obstruction by increasing the caliber of the airway. These include tonsillectomy/ adenoidectomy, tongue-hyoid suspention, glossopexy (tongue-lip adhesion), subperiosteal release of the floor of the mouth, temporomandibular joint arthroplasty, manadibular lengthening procedures, maxillary and midface osteotomies and tracheostomy.[2,4,7]

Tongue base and hyoid suspension treat the obstruction by pulling forward the base of the tongue and hyoid bone; thus expanding the upper airway. During this procedure, the superior branch of recurrent laryngeal nerve is at the risk of injury.[2-4]

Glossopexy procedure is done at the time of palatal repair near the first year of age. Glossopexy has been reported to affect the pre-speech vocalization of the child by delaying the onset of babbling and initiation of first words and causing the child to produce tongue tip sounds with the tongue blade.[8]

LeFort I osteotomy is useful for correction of retropalatal airway obstruction and can be combind with uvuloplasty and mandibular advancement. Mid-face osteotomies (LeFort III and monobloc) may be needed to correct nasopharyngeal and retropalatal airway obstruction.[9]

For most severe cases, tracheostomy is used to secure a stable airway. Tracheostomy tubes may become displaced or plugged with secretions, resulting in death. Moreover, home care after tracheostomy remains a significant burden for the family. [2,4,5]

A recessed mandible can be advanced into a normal bite through sagital osteotomy of mandibular body and ramus, in selected cases.[9] Distraction osteogenesis (DO) of the mandible offers definitive correction of micrognathia.[7] After osteotomy, DO is used to induce new bone formation between two vascularised, osteotomised bone segments which are gradually separated by a mechanical device (distractor).[3,4,6] The distractor used in the above case report was an internal, unidirectional distractor of treu dynamic brand. External distractors are difficult to look after, and they carry the risk of infection and ugly scar formation.

Generally DO consists of four consistent phases: osteotomy and device placement; latency period of primary healing from 1 to 7 days; active distraction; at a rate of 0.5-1.5 mm/day; and consolidation phase ranging from 6 to 16 weeks.

Plain radiographic evidence of bone continuity across the distraction gap is the best evidence that the device may be removed. Longer consolidation periods have been associated with increased difficulty in removing screws from internal devices and also increase the risk of pin-site infection and pin loosening.[6]

In correction of airway obstruction in patients of congenital micrognathia, neonatal MDO has yielded excellent results to provide a safe air way and avoiding tracheostomy. However, it is also associated with potential complications such as infection, tooth bud injury, inferior alveolar and facial nerve injury, poor cosmetic results, malocclusion, growth disturbance in mandible and feeding difficulties. Many of these complications can be avoided with good surgical technique and postoperative care.[10]

In conclusion, mandibular distraction osteogenesis can successfully correct micrognathia, relieve respiratory obstruction and avoid the need for tracheostomy and its associated mortality and morbidity.

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