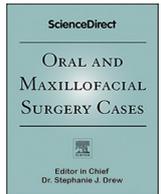




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Median cleft of lower lip with ankyloglossia: A case report

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ABSTRACT

Median cleft of the lower lip & mandible (Tessier no. 30 facial cleft) is an uncommon condition. This cleft varies in severity, in mildest form it may present as submucosal notching of lower lip & in most severe form it may involve lower lip, tongue, alveolus, mandible, floor of the mouth and neck structures. In our case report, we describe a child patient having median cleft of the lower lip with ankyloglossia, and its treatment.

1. Introduction

Clefts affecting upper lip and maxilla are common congenital entities, but midline cleft involving lower lip and mandible are rare anomalies. This cleft was first described by Couronine in 1819 [1] and since then very few cases have been reported in literature with different severity. It is also described as Cleft No. 30 in Tessier's classification of clefts [2]. This cleft may present with varying degree of severity from isolated midline cleft of the lower lip to the cleft involving lower lip, tongue, alveolus, mandible, floor of the mouth and neck structures [3,4]. We report a case of 6 months old female patient with median cleft of the lower lip & severe ankyloglossia.

2. Case report

A 6 months old female child was brought by parents to our cleft unit for correction of her lower lip deformity. On primary examination, median cleft of lower lip with ankyloglossia was diagnosed. The upper lip, nose and palate were normal. There was no known family history of cleft deformities and the pregnancy was uneventful.

On detailed physical examination, the child had median cleft of lower lip extending from vermillion to mentolabial sulcus [Fig. 1]. Intraorally she had significant ankyloglossia. The tip of the tongue was attached to alveolus [Fig. 2]. The palpation of the alveolar margin revealed small crestal notching in the midline but the mandibular surface found to be intact. Radiological examination confirmed absence of any bony cleft [Fig. 3]. Her weight & height were 5.6 kg and 57 cms respectively.

The complete workup was carried out to rule out any other anomalies. She had no other systemic abnormalities and hematological reports were within normal limit.

She was taken for the surgery under general anaesthesia. The "V" incision was placed over skin (extra orally) & mucosa (intra orally) along cleft margins. The skin, vermillion & mucosa present in cleft area were excised. The muscle was repaired first followed by closure of skin, vermillion & mucosa intra orally [Fig. 4A].

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Fig. 1. Preoperative frontal view.

The tip of the tongue was released from its attachment over alveolus & floor of mouth followed by release of lingual frenulum. The mobility of the tongue was checked by pulling it in all directions & the defect was closed [Fig. 4B].

The patient's immediate post-operative phase was uneventful. She was recalled for follow-up visits on regular intervals. During 6 months follow up lower lip appeared normal in terms of length, lips competency with good vermilion appearance [Fig. 5].

3. Discussion

The clefts involving upper lip & face are commonly seen but cleft involving lower lip and mandible are scantily documented in the literature. Only 75 to 80 cases have been reported worldwide so far, making it mandatory and worthwhile reporting to literature, whenever it occurs [5].

Several hypotheses have been proposed by different authors regarding its pathogenesis. Most of the authors consider that this anomaly occur due to failure of fusion of the first pair of branchial arches or improper mesodermal penetration into the midline of mandibular part of the first branchial arch. The theory proposed by Oostrom et in 1996 is widely accepted. According to them any hypoplasia of mandibular process during early embryonic period can lead to severe cleft of the mandible extending into the neck whereas hypoplasia during the late embryonic period will lead to less severe median cleft [6].

Therefore this deformity can vary in severity & presentation. In mild form a notching in vermilion or cleft involving lower lip only is seen. In moderate form cleft may extends into the mandibular symphysis. The tongue may be bifid & attached to the cleft alveolar margin [7]. In severe form the mandible is bifid, epiglottis or hyoid may be absent & thyroid cartilage may be underdeveloped. The midline cervical cord and strap muscles may be atrophic [8]. There may be midline dermoid in the neck, the clavicles may be spaced and manubrium sterni may be bifid [9] In our case there was median cleft of the lower lip extending up to mentolabial sulcus with severe ankyloglossia, the tip of the tongue was attached to alveolus and mild crestal notching was present in midline.

This cleft may be associated with other anomalies like congenital heart disease, hand anomalies (syndactyly and brachydactyly)

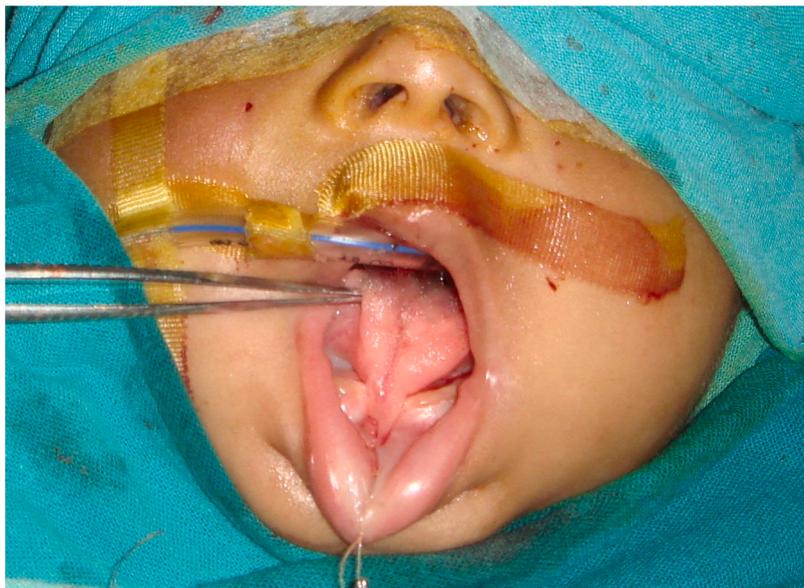


Fig. 2. Ankyloglossia.

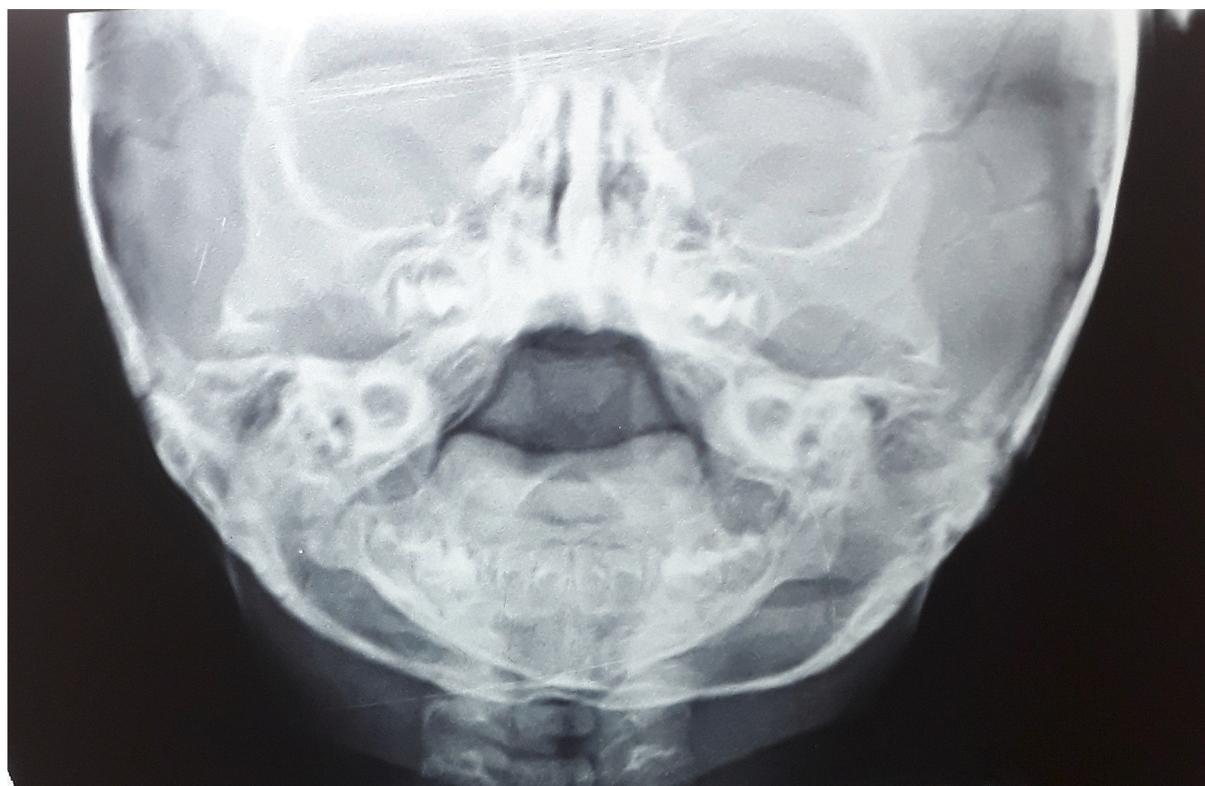


Fig. 3. Radiograph.

and foot anomalies (club foot and ectrodactyly) [10]. Our patient had no other anomalies associated with median cleft of lower lip.

The timing of surgical correction depends on severity & the structures involved. For soft tissue correction, the age-weight protocol for upper lip cleft surgery can be followed i.e. age of 6 months with having child's weight more than 5 kgs. We followed same protocol for our case and received satisfactory results. This early intervention helps child in feeding & speaking.



Fig. 4. A. Immediate postoperative, B. Release of ankyloglossia.



Fig. 5. Late postoperative frontal view.

There is lack of consensus amongst authors on the timing as well as kind of procedure for bony correction of mandibular cleft. According to Seyhan [11], if there is no gap between the two mandibular halves the bony correction can be deferred till 10 years of age, while if the patient has feeding or breathing difficulty and the mandibular segments are mobile, an early attempt should be made to stabilize the mandible with bone graft or reconstruction plate.

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Conflicts of interest

- Author 1 declares that he has no conflict of interest
- Author 2 declares that she has no conflict of interest.

Ethical approval

“All procedures performed in study involving human participants were in accordance with the ethical standards of the institutional & with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.”

Informed consent: Informed consent was obtained from the patient’s parents as the patient was minor, for procedure performed and pictures.

References

- [1] Couronne’ (of Rouen). Ann Med Montpellier 1819;107. as cited by Monroe, C. W. (1966).

- [2] Tessier P. Anatomical classification facial, cranio-facial and latero-facial clefts. *J Maxillofac Surg* 1976;4(2):69–92.
- [3] Knowles CC, Littlewood AH, Bush PG. Incomplete median cleft of the lower lip and chin with complete cleft of the mandible. *Br Dent J* 1969;127(7):337–9.
- [4] Monroe CW. Midline cleft of the lower lip, mandible and tongue with flexion contracture of the neck: case report and review of the literature. *Plast Reconstr Surg* 1966;38(4):312–9.
- [5] Rao Anantheswar YN. Complete midline cleft of lower lip, mandible, tongue, floor of mouth with neck contracture: a case report and review of literature". *Craniomaxillofacial Trauma Reconstr* 2015;8:363–9.
- [6] Oostrom CA, Vermeij-Keers C, Gilbert PM, van der Meulen JC. Median cleft of the lower lip and mandible: case reports, a new embryologic hypothesis, and subdivision. *Plast Reconstr Surg* 1996;97:313–20.
- [7] Tiwari VK. Median cleft of lower lip and Mandible: a case report. *Indian J Plast Surg* 2000;33:98–100.
- [8] Henry K, Kawamoto Jr . In: McCarthy Joseph G, Plast Surg WB, editors. Rare craniofacial clefts. Saunders Company; 1990. p. 2927–41.
- [9] Rana RE, Puri VA, Thakkur RK, Baliarsing AS. Median cleft of mandible and lower lip with ankyloglossia and ectopic minor salivary gland on tongue. *Indian J Plast Surg* 2004;37:67–70.
- [10] Ladani P, Sailer HF, Sabnis R. Tessier 30 symphyseal mandibular cleft: early simultaneous soft and hard tissue correction — a case report. *J Cranio-Maxillo-Fac Surg* 2013;41:735–9.
- [11] Seyhan T, Kilyur H. Median cleft of lower lip: report of two new cases and review of literature. *Ann Otol Rhinol Laryngol* 2002;111:217–21.