Isolated Median Cleft Tongue in the Absence of other Syndromic Characteristics: Report of a Rare Case

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Abstract

Of all the congenital defects in the oral cavity, an isolated cleft tongue is perhaps the rarest; it is more often seen along with other congenital anomalies such as cleft palate, lower lip cleft, microtia, and midline central nervous system defects. A rare case of a 35year old woman is presented, where the condition has been surgically corrected satisfactorily.

Introduction

Normally, the tongue develops during the fourth week of intrauterine life, and the fusion of the two lateral lingual swellings and the tuberculum impar, along with the mesoderm between first and the second pharyngeal arches result in the formation of the anterior two-thirds of the tongue. A cleft in the tongue may occur when this process is disturbed.

Case Report

The case of a 35-year old womanwho reported to us is presented. She complained of difficulty in cleaning her tongue, and had associated pain and halitosis. She had no known systemic illnesses. On clinical examination it was found that the median part of her tongue was covered with soft white matter, presumably food remnants and bacterial/fungal debris. After careful debridement it was clear that the patient had a deep median cleft on the whole of the anterior two thirds of her tongue. She was advised to undergo surgical correction for the same.

The cleft tongue margins were incised along the cleft borders and the unhealthy mucosa within the cleft was excised, and dissection done to expose the deeper structures such as tongue musculature. First, the muscle layers of both the sides were sutured using 3-0 absorbable sutures, and then careful approximation of the left and right parts of the tongue mucosa was completed using 4-0 absorbable sutures. She was advised to maintain good oral hygiene post operatively with 0.12% chlorhexidine gluconate mouth wash, and antibiotics prescribed to prevent chances of postoperative infection. Follow-up one week later showed that healing occurred uneventfully, and the patient had no complaints.

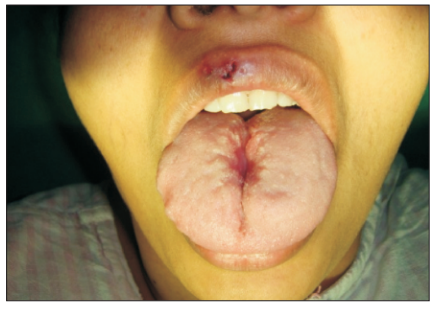

Fig 1. Initial presentation

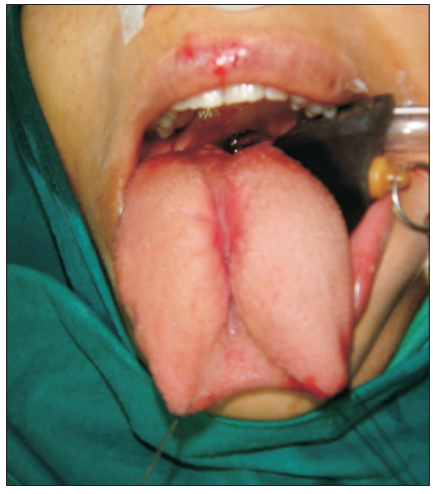

Fig 2. Post cleaning and preparation

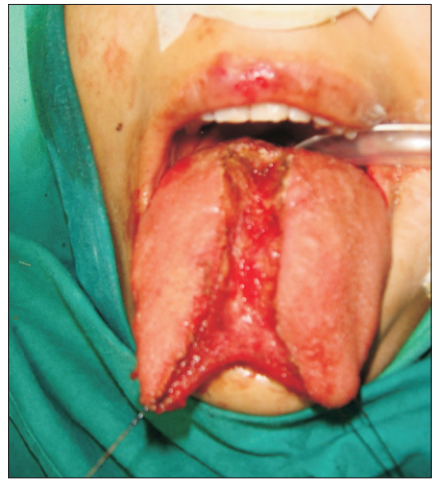

Fig 3. Freshening of wound edges

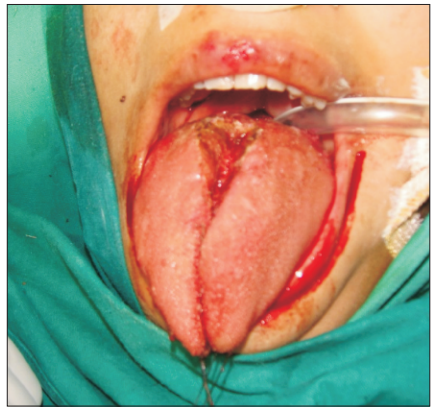

Fig 4. Approximation of the two halves

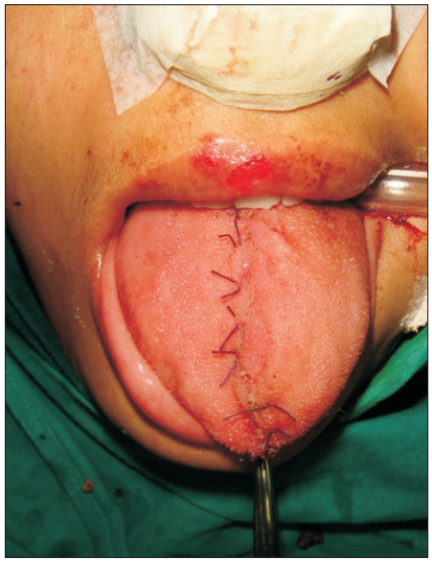

Fig 5. Closure

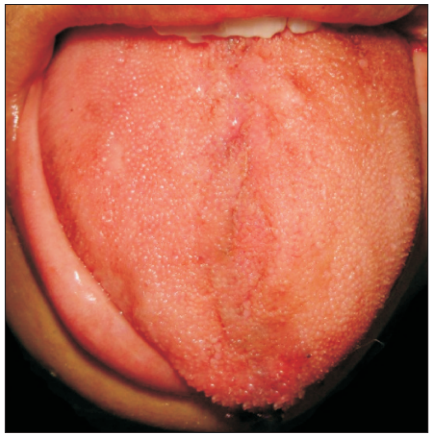

Fig 6. Post-op

Discussion

The development of the tongue starts at the fourth week of intrauterine life in the floor of the primitive cavity from the first three or four branchial arches. [1] Case reports of median cleft tongues in the literature show mostly a combination with a cleft of the median cleft lip. [2],[3],[4] Median tongue clefts are reported only to be associated with orofacial digital syndromes type I, II, IV and VI. [5],[6],[7] Bartholdsonet al described a baby boy with a bifid tongue combined with a cleft palate. [8]Bifid tongue has also been reported as a rare feature associated with infants of diabetic mother syndrome. [9] It has also been reported in syndromic cases like Opitz G BBB syndrome, Klippel-Feil anomaly and Larsen syndrome. [10],[11],[12],[13]

Abnormal/partial fusion/non-fusion of the first four arches may lead to congenital defects of the tongue, including bifid tongue. [6]Aglossia, microglossia, macroglossia, accessory tongue, long tongue and bifid tongue are the commonest occurrences listed in the order of frequency.

The treatment of such isolated cases of bifid tongue is fairly simple, with almost no complications, provided proper sterilization, isolation and surgical techniques are employed, as demonstrated in this case.

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