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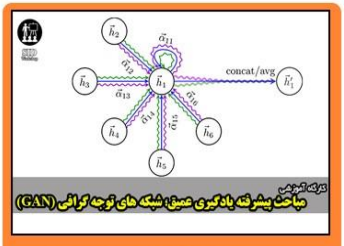

مباحث پیشرفته یادگیری عمیق؛
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Case Report

Labial Ankyloglossia Associated with Oligodontia: A Case Report

L. Chandrashekar[1]✉, K. R. Kashinath[2], Setty Suhas[3]

[1]Reader, Department of Oral Medicine and Radiology, Best Dental Science College and hospital Madurai, Tamilnadu, India
[2]Principal, Professor and Head, Department of Prosthodontics, Sri Siddhartha Dental College and Hospital, Tumkur, India
[3]Professor, Department of Oral Medicine and Radiology, Sri Siddhartha Dental College and Hospital, Tumkur, India

Abstract

Developmental disturbances of the tongue (aglossia, macroglossia, microglossia, and ankyloglossia) can adversely affect the development of the surrounding structures including the palate, alveolar process, and teeth. These developmental disturbances impair functions such as mastication, speech, and swallowing. Ankyloglossia is of various types and may be associated with other syndromic features like cleft palate. We report a rare type of non-syndromic ankyloglossia associated with missing of the permanent anterior tooth.

Key Words: Ankyloglossia; Oligodontia; Labial Frenum

✉ Corresponding author:
L. Chandrashekar, Department of Oral Medicine and Radiology, Best Dental Science College and hospital Madurai, Tamilnadu, India

drshekar75@gmail.com

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INTRODUCTION

Ankyloglossia is the restriction of tongue movement due to congenital abnormality in the lingual frenum. Ankyloglossia may be syndrome associated, but commonly it has a non-syndromic presentation with an incidence of up to 10% [1].

We present a case with an unusual type of ankyloglossia that cannot be classified using the current classification system for ankyloglossia, wherein only the tip of the tongue is fused with the labial frenum of the mandibular arch and is associated with missing of the mandibular permanent anterior teeth.

CASE REPORT

A 13-year-old girl was seen for complaining of decayed teeth. This was her first dental visit.

On examination, the mandibular right lateral incisors and bilateral central incisors were missing and the left mandibular lateral incisor was decayed. She also had generalized enamel hypoplasia and angle's class I malocclusion with bimaxillary protrusion. Her tongue was depapilated and ankylosis of the tip of the tongue with the labial frenum of the lower lip was noticed (Figure 1).

Movements of the tongue were restricted and pronunciations of consonants were affected. Family history was unremarkable. A pediatrician evaluated her general health status and she was found to be suffering from iron deficiency anemia.

After taking detailed history and complete examination, a provisional diagnosis of labial ankyloglossia associated with oligodontia and iron deficiency anemia was considered.

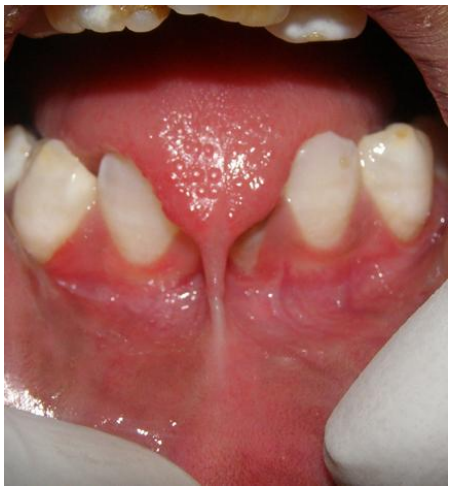

Fig 1. Fusion of tip of tongue with labial frenum

Panoramic radiography confirmed missing of the permanent mandibular central incisors and the right lateral incisor (figure 2).

After routine blood investigations, surgical release of the ankyloglossia with "Z" plasty using a 15 no. B.P blade was carried out under local anesthesia and was sutured using vicryl 3-0 suture material. Post treatment healing was uneventful and tongue movement was restored to normalcy (figure 3). Iron deficiency anemia was managed by the pediatrician uneventfully.

DISCUSSION

Treatment seeking behavior for structural birth defects with minimal or no functional deficit are generally delayed by patients, more so in the lower socio economic strata of the society. In the present case, parents of the patient had not sought any consultation for their child's tongue problem because they felt that no treatment was essential as her daily activities were not affected. Our patient presented for treatment of a decayed tooth and the presence of ankyloglossia and oligodontia were identified.

Ankyloglossia is defined as "a condition in which the tip of the tongue cannot be protruded beyond the lower incisor teeth because of short frenulum linguae, often containing scar tissue" [2]. Lower lip develops by the

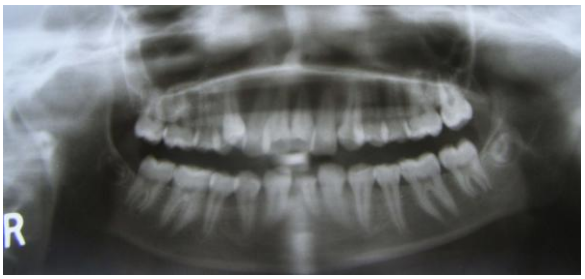

Fig 2. OPG showing missing mandibular central incisors and left lateral incisor

fusion of bilateral mandibular arches at Cernegie stage 11 of embryogenesis and it completely merges by stage 15 and the tongue tissue is found to be prominent by stage 16 [3]. In the present case, fusion of the tip of the tongue to the lower lip and missing of the permanent teeth suggest that there has been a deviation from the normal development in the region of fusion of the first brachial arches. On the review of literature, various classifications of ankyloglossia have been proposed [4]; we were unable to classify our case based on the published criteria. Incidental findings associated with ankyloglossia including median mandibular clefts [5], bifid uvula [6], undeveloped epiglottis or hyoid and underdeveloped thyroid cartilage [7] have been reported. Ankyloglossia has also been observed together with syndromes such as X-linked cleft palate syndrome [8] Kindler syndrome [9], Vander Woude syndrome10 and Opitz syndrome [9].

Labial frenum ankyloglossia has also been reported in a child associated with oligodontia of the deciduous molar dentition [10]. In the present case, ankyloglossia was associated with partial anodontia of the permanent anterior teeth and this association has not been reported previously. Presence of iron deficiency anemia in the present case could be another non related finding. Hanhart syndrome and holoprosencephaly or microforms of holoprosencephaly have been associated with tongue anomalies; however, other features like skeletal abnormalities of the midface, limbs and CNS anomalies are also part of the spectrum

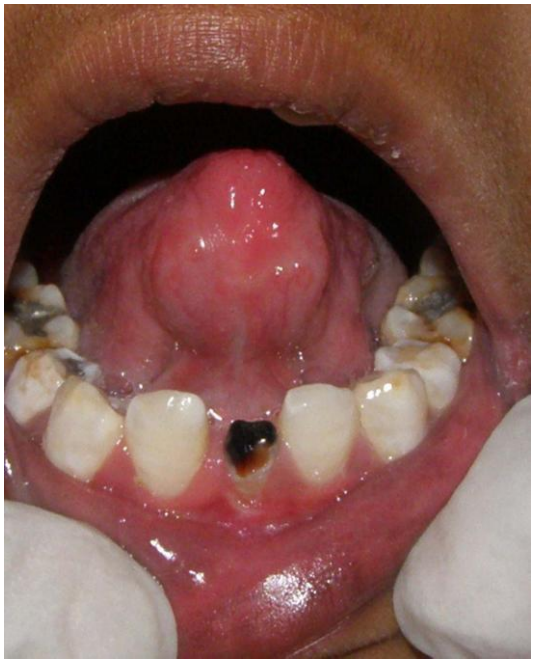

Fig 3 . Tongue after surgical release

of other anomalies in these patients [11, 12]. Abnormal development of the palate (cleft palate) and tongue (ankyloglossia) have been associated with aberrations in T-bx22 genes. Mutations in this gene have been associated with cleft palate and ankyloglossia, and it is believed to play a major role in human palatogenesis [13]. Preferential tooth agenesis (incisors and/or premolars) associated with orofacial clefts have been identified in Interferon Regulatory Factor 6 (IRF6) gene deletions and point mutations [14]. As there was no cleft palate in our case, we can draw an inference of the possibility of absence of this mutation.

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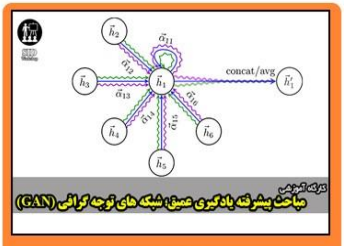

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