Case Report

Accessory oral cavity

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INTRODUCTION

The presence of accessory oral cavity is very rare on review of literature, though references for teratoma and sublingual dermoid cyst and accessory tongue individually seen are plenty. The patient was a single child for the parents of non-consanguineous marriage born of normal delivery and no other problems. The reason for the presence of such an anomaly could not be drawn.

CASE REPORT

A female patient aged 11 years had reported to the hospital with her parents with complaint of presence of extra mouth since birth. They were embarrassed and had not been treated well by others in the village, thought it to be a bad omen, and came to remove it. There were no other specific problems due to the extra mouth. The patient was brushing that accessory mouth.

On examination there were two oral cavities, one normal and other accessory.

Normal oral cavity
On extra oral examination of the normal oral cavity, the upper and lower lips were normal. The maxilla was normal. The mandible was asymmetrical due to the bony prominence.

On intra oral examination the buccal and labial vestibules of the maxilla were normal. In mandible, the buccal vestibule was normal, but the depth of the labial vestibule was obliterated due to the prominent presence of the accessory oral cavity. The maxillary and mandibular posterior teeth were in normal occlusion, but the mandibular anterior teeth were present at the right side were inclined lingually, but in the left side lower anterior teeth were normally placed.

The position was elevated at the right side due to the presence of the cystic lesion of the floor of the mouth, which extends from the midline to right retromolar region. But there was no disturbance in normal swallowing. The tongue movement was normal. Speech was normal.

ABSTRACT

This is a rare case report of a patient around 11 years with the complaint of extra mouth who reported to the hospital for removal of that extra mouth. On examination there was accessory oral cavity with small upper and lower lips, seven teeth and saliva was drooling out. Under general anesthesia crevicular incision from 32 to 43 was put and labial gingiva with alveolar mucosa was reflected completely and bone exposed to lower border of mandible. There were seven teeth resembling lower permanent anterior teeth in the accessory mouth, which was excised with the accessory lips. 41 extracted and osteotomy carried out extending the incision from the extracted site and osteotomy carried out. Dermoid cyst both below and above the mylohyoid muscle and rudimentary tongue found and excised and the specimen sent for histopathological examination. The wound was closed and uneventful healing noted to the satisfaction of the patient. This is a rare and interesting case which has been documented.

Key words: Accessory mouth, accessory oral cavity, osteotomy, sublingual dermoid, teratoma
Accessory oral cavity
On extra oral examination, the accessory oral cavity was situated below the lower lip of the normal oral cavity on the right symphysis region. There were small upper and lower lips fused at the angle. The right angle was situated 1 cm below the right angle of the normal oral cavity and the left angle was crossing the normal midline extending ½ cm on the left symphysis region. There was skin present in between the lower lip of the normal oral cavity and the accessory upper lip.

On intra oral examination in the center of the accessory oral cavity there was a bony prominence with the presence of seven lower anterior teeth with gingival attachment. The base of the bony prominence was fused with the labial surface of the mandible of normal oral cavity. There were associated right and left buccal and labial vestibules. Salivary discharge too found.

The patient was not able to swallow through that accessory mouth and was able to move both the upper and lower lips of that accessory oral cavity and able to approximate both the lips of the same [Figures 1-3].

CT Examination shows excessive growth of mandible with presence of cystic swelling and radiolucency on the lingual surface of mandible extending from the midline to right angle of mandible [Figures 4-6].

3D CT shows excessive bony growth over the labial surface of the mandible [Figure 7].

Procedure
Under nassoendotracheal intubation first crevicular incision from 32 to 43 was put and labial gingiva with alveolar mucosa was reflected completely and bone exposed to lower border of mandible. During the procedure labial mucosa attached to the bony prominence over accessory mouth was incised and bony prominence was brought to normal oral cavity.

There were seven teeth resembling lower permanent anterior teeth. Then excision of accessory lips was done
around the vermillion border. Inside the oral cavity 41 was extracted and through that socket osteotomy done. Vertical incisions were put over 41 extracted site and between 43 and 44 and horizontally connected half cm below the lower border of bony prominence of accessory mouth. The lesion was displaced lingually and mylohyoid muscle exposed and able to see dermoid cyst both below and above the mylohyoid muscle. Through the same approach excised the entire dermoid cyst. During the excision could see one
small rudimentary tongue approximately 1-1/2 × 1 cm which was attached with the lining mucosa of dermoid cyst. There were multiple calcified granules and hair follicles filled with straw colored fluid. This dermoid cyst was attached on the lingual surface of the excised accessory bony prominence and there was communication from the dermoid cyst to the oral cavity. There was separate blood supply to the mouth.

The excised specimen was sent for biopsy. Closure was done intraorally with 3-0 chromic catgut and normal labial lower vestibule was reconstructed. The outer skin layer was approximated with lower portion of the skin of original lower lips with 3-0 black silk. The postoperative course was uneventful and the patient went home happy [Figures 8-12].

HPE
Revealed teratoma with presence of multiple small bony granules and hair follicles and teeth. The rudimentary tongue too was confirmed by the pathologist [Figures 13-15].

**DISCUSSION**

**Accessory mouth**
Al–Wahedi Em in 1995 documented one rare case of teratoma with a double mandible and double mouth, mucous secretions and two teeth erupting in the accessory mouth. There were numerous supernumerary teeth in the main body of the mandible, and gerninated primary teeth in the mandibular arch. There was a history of cleft palate. Other than this no other
references for accessory oral cavity could be found which is to be mentioned.[1]

Review of literature only shows teratoma, sublingual dermoid and accessory tongue as an individual entity unlike in this case where it is totally a new accessory oral cavity.

**Teratoma**

Teratomas are true neoplasms composed of tissues of all three germinal layers. They have an unknown origin and eccentric microscopic appearance. Around 80% are located in the ovaries and sacral lesions, 7% are seen in head and neck region, only approximate 1.6% of these tumours are found in oral region. Teratomas arising from the oral cavity are rare in the newborn. It is a benign tumor, although malignancy has been described in adults. In utero it can cause polyhydramnios or fetal death. In the newborn it can cause respiratory distress due to tracheal obstruction. Head and neck occurrence is usually localized to the neck and nasopharynx and comprise 1-10% of cases. Other extracervical presentation of teratoma is very rare. The histogenesis of teratoma remains debatable.[2] Pure oral presentation in the tongue is extremely rare which was reported once by Gupta S et al.[3] Also there is a report of an oral teratoma - a mouth tumor - was successfully removed from a fetus while still in the womb via operative fetoscopy, doctors from Jackson Memorial Hospital, Florida, reported in the American Journal of Obstetrics and Gynecology. The medical team says this procedure was the “world first”.[4]

**Dermoid cyst**

Dermoid cysts occur primarily in sites where embryonic parts fuse together.[5] The majority of reported cases are in the midline of the body and especially in testis and ovaries and in the head and neck region. Dermoid cysts may be classified into two major categories: congenital and acquired forms. Congenital dermoid cysts arise from epithelial rests trapped during midline fusion of these branchial arches whereas acquired dermoid cyst arise from epithelium implanted during trauma and they occur at the sites away from midline. Dermoid cysts in the floor of the mouth may be congenital or acquired. The congenital form, according to the main theory, originates from embryonic cells of the 1st and 2nd branchial arch. The acquired form may be due to traumatic or iatrogenic causes and as a result of the occlusion of a sebaceous gland duct. They may also be classified as anatomical and histological. Anatomically, they are divided into median genioglossal, median geniohyoid, and lateral cysts, while histologically they are divided into epidermoid, dermoid cysts and teratomas. Clinically, a distinction between supra and inferior type as well as between central and lateral type is proposed in relation to the mylohyoid muscle and the midline, respectively. Histologically, an estimation of dermoid, epidermoid, and teratoid cysts is reported. In international bibliography, reference is made to three theories with regard to the origin of cysts in the floor of the mouth.[6-14]

Enucleation via intraoral and/or extraoral approach is the method of treatment.

**Tongue**

The development of the tongue begins as known, in the floor of the primitive oral cavity, when the human embryo is four weeks old. More specifically, the tongue develops from the region of the first three or four branchial arches during the period that the external face develops. Malformations of the tongue are structural defects, present at birth and happening during embryogenesis.[13] One of the most common malformations is accessory tongue which has been reported in literature. Cases of accessory tongue as a separate entity has been documented and found in few articles.[16-18]

**Conclusion**

This rare case was only case once reported in review of literature and hence found to be interesting and documented.

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