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Case Report

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A rare case report of Congenital Glossocervical Fistula

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Abstract

The vast majority of all branchial disorders are thought to be associated with the second branchial cleft apparatus. Lateral cervical fistula results due to incomplete obliteration of second branchial cleft and pouch. Classically these fistulae have a well-defined pathway through the neck. Most common opening in the oropharynx is the tonsillar fossa. An incomplete branchial fistula is a common anomaly but complete branchial fistula is rare. Here we report a case of a left glossocervical fistula with external opening over anterior border of sternomastoid muscle passing deep to platysma with internal opening at lateral part of base tongue at the level of circumvallate papillae which is very rare.

Keywords: Branchial fistula, Second arch, Tonsillar fossa, Glossocervical fistula.

Introduction

The branchial cyst, sinus and fistula are anomalies of branchial apparatus which consists of five mesodermal arches separated by invagination of ectoderm (branchial cleft) and endoderm (branchial pouch). The branchial fistula is not a true fistula because it rarely has two openings.[1] More than 90% branchial cleft anomalies arise from second branchial cleft system and 8% first branchial cleft system. Anomalies from third and fourth branchial cleft system rarely occurs.[2]

In the embryo the second arch grows caudally to cover third and fourth arches and second, third and fourth clefts, eventually fusing with lower neck. The buried cleft persists as cavities by endoderm and generally disappears with development. If this does not occur, it persists as branchial cyst. Failure of this obliterative process and the formation of an opening between the pouch and cleft are considered to be responsible for congenital fistulae between the oropharynx and lateral neck.[3]

A 15-year old female patient presented to ENT OPD, with the complaint of a small opening on upper part of neck on left side since birth and watery discharge from the opening while drinking.

The general examination was within normal limits. On examination of neck a small opening was seen at the level of hyoid bone on anterior border of left sternocleidomastoid muscle (Figure 1).

On palpation thick cord like structure was felt which extended upto angle of mandible. No internal opening was seen on naked eye examination of oropharynx. On the basis of clinical findings she was diagnosed a case of congenital branchial fistula. To confirm the diagnosis fistulogram was carried out. The fistula showed a tract which was dilated

in its upper one-third (Figure 2) and it ended in the oropharynx.

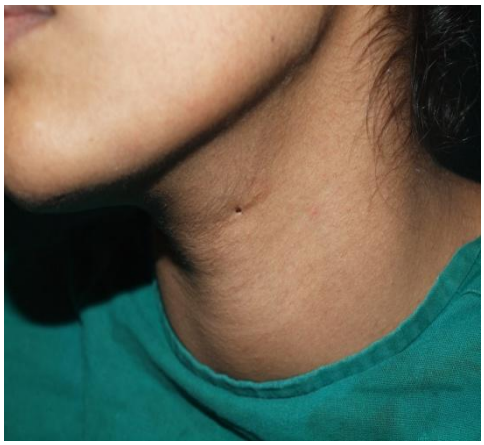

Figure 1: Small opening seen at the level of hyoid bone on anterior border of left sternocleidomastoid muscle

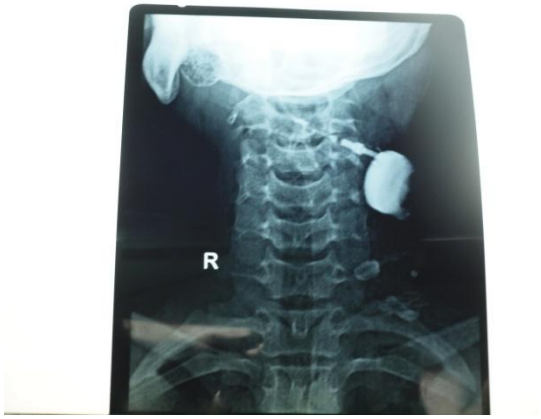

Figure 2: Fistulogram showing a tract

The radiologist reported a free flow of dye in oropharynx during procedure indicating that there is a free communication with oropharynx. CT scan with contrast was taken which showed the path of the tract, which was opening at the base of tongue (Figure 3 and 4).

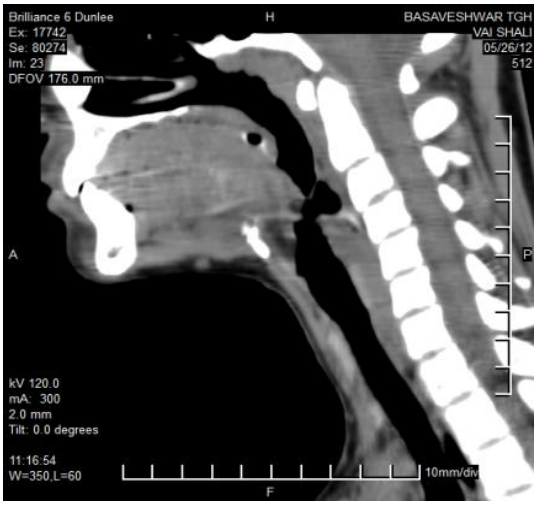

Figure 3: CT scan of the oropharynx

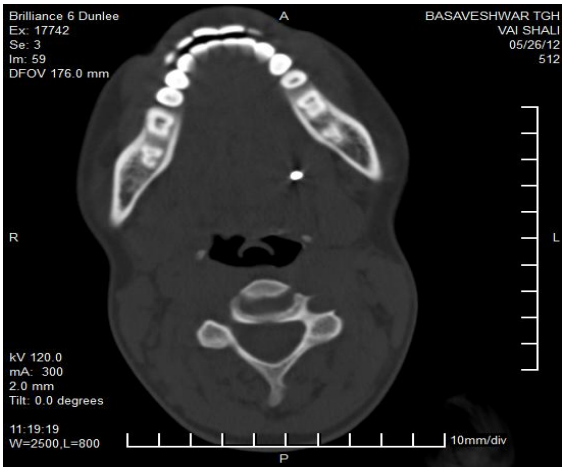

Figure 4: CT scan of the oropharynx

After necessary investigation for fitness for general anaesthesia excision of fistulous tract was carried out through external approach. An elliptical incision was given around the external opening and subplatysmal flaps were raised. The fistulous tract was infused with methylene blue dye. The tract was identified as thick cord like structure which was dissected in subplatysmal plane upto the base of tongue. Prolene 0 had been passed through the fistulous tract at external opening which came out through an internal opening in oropharynx (Figure 5) at the base of the tongue which was identified endoscopically (Figure 6).

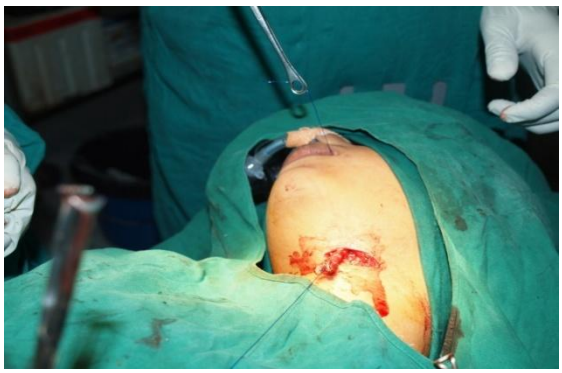

Figure 5: Fistulous tract opening in oropharynx

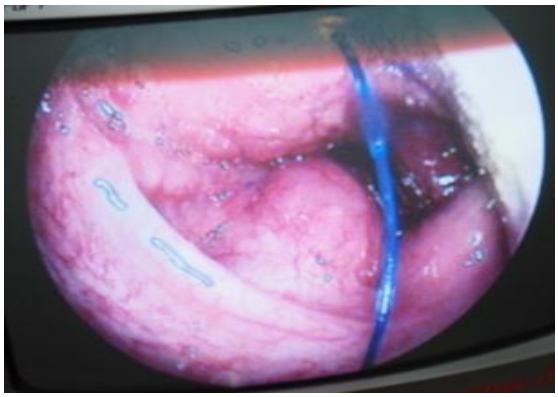

Figure 6: Fistulous tract opening in oropharynx identified endoscopically

The tract was seen to be passing between external and internal carotid artery, tracing upward below the stylohoid ligament. Fistulous tract had been excised (Figure 7 and 8) which measured around 7.5cm (Figure 9). Base of tongue defect sutured with vicryl 3-0. Complete haemostasis was achieved and neck wound closed in layer. The tract was sent for HPE. Microscopic examination of specimen revealed a linear tract lined by keratinized stratified squamous epithelium. In the wall of tract clusters of mucous secreting glands were seen.

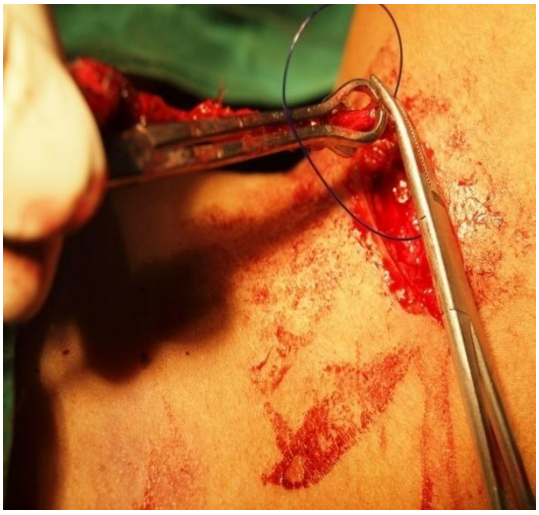

Figure 7: Excised Fistulous tract

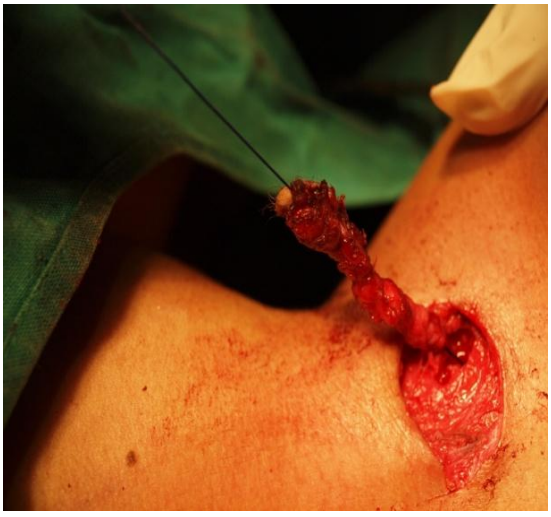

Figure 8: Excised Fistulous tract

Base of tongue defect sutured with vicryl 3-0. Complete haemostasis was achieved and neck wound closed in layer. The tract was sent for HPE. Microscopic examination of specimen revealed a linear tract lined by keratinized stratified squamous epithelium. In the wall of tract clusters of mucous secreting glands were seen.

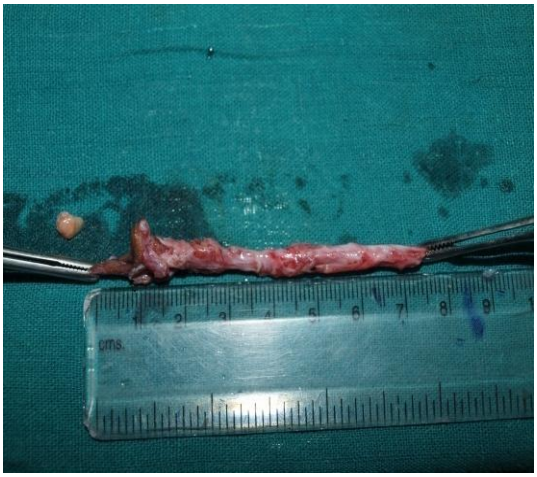

Figure 9: Excised Fistulous tract meauring around 7.5cm

Discussion

The branchial cyst and sinus are more common in male1 (60%).Peak age for presentation of cyst is in the third decade and that of the congenital sinus at birth. Acquired sinuses are produced due to infection of branchial cyst and their rupture. More than 90% of branchial cleft anomalies arise from second branchial cleft system and 8% from the first branchial cleft system.[4]

The branchial sinus and fistula present in the neonatal period with an external opening along the line joining the tragus and the sternoclaviclar joint along the anterior border of sternocleidomastoid muscle. They are mostly found on the left side of neck. To understand the extent and course of the branchial cyst, sinus and fistula, it is essential to known the embryologic development and related anatomy.[5] These anomalies may originate from the first to fourth cleft/pouch, with the commonest (95%) arising from the second cleft/pouch.[6] Persistence of ectoderm in the fused tube gives rise to branchial cyst. The branchial fistula results from the breakdown of the endoderm, usually in the second pouch.[7]

The first branchial arch anomalies are classified by work (1997) as types I and II. Types I branchial defects are duplication anomalies of external auditory canal which exists as fistulous tract near the lower portion of parotid gland in close association with the facial nerve. They present as sinus tracts near the postauricular sulcus or concha or anterior to the tragus. These anomalies course through the infratemporal fossa parallel to EAC and may end either in the EAC or middle ear space.

Types II defect is less common and presents as a cyst or sinus in the anterior triangle of neck below the angle of mandible. These anomalies course superiorly and

posteriorly through the parotid gland in close proximity of facial nerve and terminate laterally in the region of bony cartilaginous junction of EAC. Second branchial anomalies present along the anterior border of sternocleidomastoid in its lower third. A tract commonly extends into the pharynx entering anywhere from nasopharynx to hypopharynx but most commonly in the region of tonsillar fossa. The tract usually passes between the second and third arch structures, i.e. it passes medially between the internal and external carotid arteries above the glossopharyngeal nerve and below the stylohyoid ligament.

Third branchial anomalies present along the anterior border of the sternocleidomastoid muscle in the lower third of the neck. The tract passes behind the internal and external carotid arteries and the glossopharyngeal nerve and while crossing over the hypoglossal and superior laryngeal nerves. It enters the pharynx at the level of the pyriform fossa. Fourth branchial anomalies are theoretically possible. These anomalies present as sinus tracts in the anterior triangle in a fashion similar to that of second and third branchial anomalies. If they exist their tract passes below the arteries of the fourth arches into the mediastinum and there continues superiorly along the ascending portion of recurrent laryngeal nerve to enter the upper part of oesophagus.

In our patient, the branchial fistula was complete. The external opening was in upper part of neck present since birth. For investigating complete branchial fistula, a fistulogram can be performed to completely delineate the tract. During excision of the tract in this patient there was no mesodermal tissue separating external and internal part. The internal opening was located over the lateral part of base tongue at the level of circumvallate papillae. These characteristics make this case a rare variety of branchial fistula. Only one such similar case has been reported in literature by Martin J. Donnelly et al in year 1994 in Dublin.[8] The recurrence rate of branchial anomaly is 3% for a primary lesion and as high as 22% for lesions with previous infection and surgery.[9] These fistulae take a well-defined pathway through the structures of the neck.[10]

Conclusion

We report a case of a left glossocervical fistula with external opening over anterior border of sternomastoid muscle passing deep to platysma with internal opening at lateral part of base tongue at the level of circumvallate papillae which is very rare.

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