Accessory parotid gland with ectopic fistulous duct - Surgical view: A case report and review of current literature

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ABSTRACT

Accessory parotid glands are a common clinical occurrence and usually drain into the main Stenson’s duct by small ductules and thereby, into the buccal cavity. Presence of an accessory parotid gland with an ectopic fistulous duct is a rare occurrence. Clinical findings, imaging studies, biochemical tests, histopathological examination are needed for appropriate surgical management. It is extremely rare case with ectopic fistulous duct in an accessory parotid gland managed surgically by internalization of the duct to open into the buccal mucosa and excision of pre-aural appendages. Further to this, we give a comprehensive review of literature on accessory parotid gland and duct anomalies.

Key words: Accessory parotid gland, congenital parotid fistula, ectopic fistulous duct, pre-aural appendage

INTRODUCTION

Most parotid fistulas are acquired and arise secondary to trauma, surgery, malignant tumors and inflammation. Rarely do they occur congenitally as malformation during fetal development. Although accessory parotid gland is a common variation, parotid fistula arising from an ectopic accessory parotid gland is extremely rare. Only five cases have been reported world-wide, so far excluding the present case.

We report a case of congenital parotid fistula arising from an ectopic accessory parotid gland (right) demonstrated by computed tomography (CT), sialography and CT fistulography.

CASE REPORT

A 10-year-old male child presented with history of intermittent discharge of watery fluid from an opening in the right cheek since birth. The discharge increased on feeding/eating and was not associated with any swelling or pain. There was no previous history of swelling, redness, pain, or injury to the region. He was otherwise healthy and had normal developmental milestones.

Patient had normal general/systemic examination with vital parameters. Local examination revealed a small punctum on the right side of the face, at a distance of about 2.5 cm from the angle of the mouth and 1 cm inferior to a line joining the tragus and angle of mouth [Figure 1a]. Expression of clear serous fluid on local massage was seen. No associated palpable mass or tenderness was noted. Oral examination was normal with normal Stenson’s ducts opening bilaterally.

Bilateral pre-aural appendages were noted on regional examination, two on the right side anterior and anteroinferior to the tragus and one appendage on the left side just anterior to the tragus [Figure 1a and b].
As a part of the diagnostic work-up, ultrasonography, sono-fistulography, contrast digital fistulography, contrast digital sialography and contrast CT fistulography [Figure 2] were done. Laboratory analysis of the fluid revealed raised salivary amylase levels (7800 IU/mL), which confirmed the diagnosis of a salivary fistula. Contrast fistulography revealed an opacified tubular shaped density visualized with opacification of soft-tissue area noted on the plain image. No intra oral spillage of contrast was seen.

Our patient was successfully managed by a simple surgical technique, described below:

The procedure was performed under general anesthesia with local infiltration of 1 in 100,000 adrenaline around the fistulous opening to minimize intra-operative bleeding. Methylene blue was then injected into the fistulous opening using a 26-gauge needle (blunt tip) under microscopic magnification. An incision-along skin crease enclosing fistulous opening was given. The duct was dissected for about 1.5 cm. Then brought out to the mucosal surface through buccinator muscle opposite the first upper molar tooth. The duct was secured in place using absorbable sutures. Skin incision was closed. Pre-auricular tags were also excised. Figures 3 and 4 are showing the dissection.

Post-operatively, patient was kept on nil by mouth for 24 h and put on intravenous fluids, antibiotics, atropine and analgesics. On stimulating the accessory gland the saliva was found dribbling from the transposed duct opening. Patient was discharged on oral antibiotics and analgesics on the 3rd post-operative day. Her sutures were removed on the 6th day. Histopathological examination of the tissue/fluid adjacent to fistulous tract showed no underlying malignancy or evidence of any specific (granulomatous) disease. At 2 weeks later sialogram was carried out which confirmed the patency of the duct. Our patient was followed-up 3 months later and was found to have successful healing of her wound with no complications or recurrence. Figures 5 and 6 are showing the post-operative status.

**DISCUSSION**

Parotid glands are paired serous salivary glands located lateral and posterior to the masseter muscle. They extend from the zygoma superiorly, to the angle of the mandible inferiorly and are limited posteriorly by the external auditory meatus. Anteriorly, the gland extends to near half of the masseter width. The facial nerve divides the parotid into the superficial and deep lobes. The gland is drained by the main parotid duct or Stenson’s duct into the buccal cavity. The Stenson’s duct arises from the anterior margin of the gland, courses anteriorly, slightly
Accessory parotid glands have been described in about 21% of general population. These glands, mostly pea sized, unilateral, are located superior to the main parotid duct, anterior to the masseter and separate from the parotid gland. They drain through one or two channels into the main parotid duct. They may consist of both serous and mucinous acini.

Frommer first described the incidence of accessory parotid gland as 21% in a study of 96 human cadavers. However, no record exists of the incidence of accessory gland with an ectopic duct. Accessory parotid gland having a duct of its own is exceedingly rare. The duct courses anteriorly, inferiorly and opens externally to the skin behind the angle of the mouth. An association has been observed between co-existent pre-aural skin appendages. However, this association is yet to be proven conclusively as very few cases have been reported until date.

Naguru and Miyazawa reported the first case of salivary fistula associated with aural appendage in 1972 from Japan. Yamasaki et al. used the Delore’s method of surgical translocation of the ectopic fistulous duct to the buccal cavity in a similar case in 1986. Moon et al. reported a case in a 5-year-old girl with an ectopic accessory parotid gland with a fistulous duct from Korea. They accurately demonstrated the ectopic gland by CT sialography and CT fistulography. However, they were unable to demonstrate the normal ipsilateral parotid gland, thereby, justifying the use of their term ectopic accessory parotid gland.

Gadodia et al. in their study have reported the imaging findings in an 8-year-old boy with congenital salivary fistula from an accessory parotid gland in 2008, from India.

Hah et al. reported a surgical management method for a similar case, also from Korea by chemo cauterization using tri-chloro-acetic acid (TCA) in 2008. They suggested that most surgical incisions for operating fistulae in the face leave a scar; hence chemo cauterization by TCA is a better option. However, in our case, we would like to point that if the accessory parotid gland has no other drainage tract then it would be unwise to cauterize the fistula leaving the gland intact, which can get infected later on. If cauterization is deemed necessary then the accessory gland must also be excised. A better option would be to internalize the ectopic duct opening and leaving the accessory gland in situ to drain into the mouth.

In our case, the 10-year-old male child presented with a fistula on the right cheek with bilateral pre-auricular

inferiorly and superficial to the masseter muscle where it is palpable, then turns medially at the anterior margin of masseter to pierce the buccinator muscle and open into the mouth. The internal opening is a slightly elevated punctum at the level of the upper second molar tooth.
appendages. Acquired causes such as traumatic, infective, or post-surgical could be ruled out at the outset as history was from birth. Imaging work-up findings along with history and examination findings of serous discharge on massage and eating proved that the fistulous duct originated from an accessory right parotid gland.

“Accessory parotid gland with ectopic fistulous duct” would be the appropriate nomenclature, as there was a normal parotid gland with its normal duct and an accessory gland with an ectopic duct which did not communicate with the Stenson’s duct, but opened as a fistula over the cheek.

Fistulae in the parotid region generally arise secondary to trauma, surgery, malignancy, or infection and are seen more often in adults.[8] They can be differentiated from congenital fistulae based on history. Fistulae in pediatric age are mostly congenital. The congenital fistulae in the region of the face and neck result from the partial failure in normal involution of the branchial arches during embryonic development. Important differential would be pre-auricular sinuses, which are seen more posteriorly near the tragus, end blindly and may be visualized on ultrasound. Local lesions like epidermal inclusion cysts or basal cell carcinoma which can present as discharging sinus can be excluded by clinical examination and histology.

The main parotid duct marks the landmark between the closing mandibular process and the maxillary process of the face during embryogenesis. The parotid gland can be recognized in human embryos at Stage 15 as an elongated furrow running dorsally from the angle of the mouth between the mandibular and maxillary prominences. The groove, which is converted into a tube, loses its connection with the epithelium of the mouth except at its ventral end and grows dorsally into the substance of the cheek. The tube persists as the parotid duct and its blind end proliferates in the local mesenchyme to form the gland. Subsequently, the size of the oral fissure is reduced by partial fusion between the maxillary and mandibular prominences and the duct opens thereafter on the inside of the cheek at some distance from the angle of the mouth. The parenchymal part of the parotid gland is formed by branching of the blind end of the parotid tube and by the development of structures of ectodermal (secretory parenchymal) and mesodermal (stromal) nature. The accessory parotid is derived from a similar branching and a similar glandular proliferation, arising from the parotid tube more anteriorly and clearly separate from the main parotid tissue.[9,10]

However, in our case we postulate that around the same time as the main duct develops from the furrow between the maxillary and mandibular processes, at about 4 weeks of development, the primeval groove duplicates and grows cranially and posteriorly to form two ducts and subsequently, the normal and accessory parotid separately. During fusion of the maxillary and mandibular processes, the normal parotid duct remains patent from inside the mouth, whereas the duct of the accessory gland opens as a cutaneous fistula.

The auricle is formed from six hillocks, out of which the tragal hillock along with two other arises from the first branchial arch. Pre-aural appendages are remnants of additional tragal hillocks.

Thus, co-existence of accessory parotid gland with ectopic fistulous duct and pre-aural appendages can be explained as an example of malformations of the first branchial arch derivatives.

Excluding our case, only five such cases have been reported in English literature and all from Asian countries. As a result, the exact nomenclature of this condition is yet to be concluded upon. There could be a regional or racial predilection to this condition. Unless a substantial number of cases are studied, the epidemiology and pathogenesis remain controversial.

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