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Case Report

First Brachial Cleft Cyst and Sinus: A Rare Presentation

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Abstract

Branchial cleft cysts, also known as Branchial cleft anomalies, are congenital anomalies arising from the first through fourth pharyngeal pouches. These anomalies can exist as true fistulae, cysts, or sinus tracts, depending on the degree of incomplete obliteration during embryogenesis. A 16year old male presented with complaints of intermittent discharge clear fluid and then pus discharge from the right post auricular sinus. Previously patient had undergone incision and drainage twice for the cystic swelling at the same site. Hrct and sinogram were inconclusive. Patient was taken up for surgical excision and exploration during the surgery tract was found leading into the parotid tissue. We classify this case as work's type 1 due its location post auricular region and relation to parotid gland.

Keywords: First Brachial; Sinus; Cleft Cyst; Anomalies

Introduction

First Branchial cleft anomalies are thought to originate from the Branchial apparatus that did not completely obliterate during head and neck embryogenesis. The incidence is estimated to be about one per million population/year [1,2]. Branchial cleft anomalies present in one of three forms: cysts, sinuses, or fistulae. First Branchial cleft anomalies are rare accounting afor < 8% of all Branchial anomalies [1]. The usual site of presentation is in the lateral neck below the external ear canals, above the hyoid bones. They are twice more common in women as compared to men with the left side predominance for fistulas [3].

Here, we report an extremely rare variant of first Branchial cleft anomaly presenting as sinus opening near mastoid tip in a young male patient.

Case Report

A 16-year-old male patient presented with complaints of right post auricular swelling and pus discharge (intermittent) from behind the right ear since 1 year. No history of ear discharge, hearing impairment, tinnitus, vertigo. History of incision and drainage present in the past twice at another clinic. Hrct temporal bone revealed no significant abnormality. Patient undergone excision under GA in March 2021, but recurrence occurred after about a month, in the form of a post auricular swelling which got burst and started discharging. On examination post auricular curvilinear scar was found in the groove with sinus punctum visualized in the post auricular scar region near the mastoid tip and the yellowish pus discharge was present. patient was given antibiotics and local ointment for application. When the discharge got dried up patient was sent for sinogram but the dye regurgitated and was inconclusive.

Patient was taken up for revision excision (Figure 1) and exploration in July 2021 under general anaesthesia. Elliptical incision was taken over seared wound. Blunt and sharp dissection done. Tract identified (Figure 2) and dissection done along the tract. Tract was visualized further into the parotid gland dissected till the external auditory canal when surgical knot taken and cut, the end was then cauterized. Haemostatic achieved, betadine wash given and suturing done. Sutures were removed on 7th postoperative day (Figure 3). The tract was sent for histopathological examination.

Figure 1: Pre surgical photograph showing post auricular scar and sinus opening.

Figure 2: Tract identified during the surgery.

Figure 3: Post surgery after suture removal.

Histopathological examination report stated stratified squamous epithelium and sub epithelial fibro collagenous tissue showing chronic inflammatory cell infiltrates mainly lymphocytes and macrophages along with few dilated and congested blood vessels. No atypia, malignancy or granuloma is seen in sections examined (impression- chronic nonspecific granulation tissue).

Discussion

Anomalies of first brachial cleft arise due to incomplete closure of the ectodermal portion of the cleft. The formation of fistula, sinus or a cyst depends on the degree of the closure of the tract. The anomalies start from the floor of the External auditory canal either at the bony cartilaginous junction or on the cartilagenous part follow the tract between the mandibular and hyoid arches and ends in the submandibular area. These lesions are located in close proximity of the parotid gland especially the superficial lobe which overlies the lesion. Very often the diagnosis of these lesions can only be made postoperatively [3].

A first Branchial cleft cyst is typically smooth, non-tender, fluctuant mass found between the external auditory canal and submandibular area. Frequently, it will have a cutaneous punctum from which fluid may be expressed. It is variably involved with the parotid gland and facial nerve, and there may be a connection to the middle or external ear, so an otologic exam is crucial in these patients [5].

There are two main types of first Branchial cleft anomalies - type-1 and type-2 on the basis of anatomical location proposed by Arnot (1971) and both location and histology by work (1972) (Table 1). Arnot's type-1 lesions included cysts or sinuses in the parotid gland, whereas type-2 were found between upper neck and external auditory canal (EAC) [6]. Work's type 1 lesions were adjacent to the EAC, whereas type 2 was in the upper neck near the angle of the mandible [6,7]. Type-1 lesions of work occur medial to concha and frequently extend to the post-auricular crease or supra-auricular region [8,9]. We classified our case into work's type-1 based on the anatomical location and its relationship to parotid gland. Another classification is given by Olsen in 1980 who proposed a simpler classification system based on the clinical presentation of the lesion: like Type 1 is a Cyst, Type 2 is a Sinus and Type 3 is a Fistula. These classification systems are debatable and lesions often are difficult to put in a particular category even after complete investigations especially in Works classification which can only be used retrospectively [10-12] as once the lesion has been excised and examined. Moreover these classifications do not help in any way in the management of these lesions.

Classification	Type 1	Type 2
Arnot classification (1971)	Cysts or sinuses in the parotid gland which are lined by squamous epithelium Appear during early or middle life Occur due to cell rests buried during obliteration of first brachial groove	Cyst or sinus in anterior cervical triangle with a communicating tract to EAC Develop during childhood Occur due to incomplete closure of the cleft
Work classification (1972)	Rare Purely ecto-dermal Duplication of membranous EAC Cysts lined by squamous epithelium, form keratin. Usually located posterior to pinna and/or medial concha with bulging into the external auditory meatus. A sinus develops after rupture or secondary infection. They may have a tract running anterior and deep to the ear lobe, parallel and medial to normal EAC superior to the main trunk of facial nerve, and finally end in a cul-de-sac on a bony plate of EAC at the level of mesotympanum	More common Ecto--dermal and meso-dermal Duplication of membranous and cartilaginous portions of EAC Cysts, sinuses or fistula. Intra-meatal p-art of work's type 2 anomalies show less protrusion of the intra-meatal part than type 1 lesions because in type 2 lesions this part is lined with cartilage Associated with a sinus/fistula opening in the region of upper neck, extend superiorly through the parotid gland towards the floor of EAC at the bony-cartilagenous junction or the cartilaginous portion. Type 2 lesions are medial or lateral to the facial nerve in the parotid gland and they might cross several branches of the facial nerve

Table 1: Classification of first Branchial cleft anomalies.

During the surgery we found the tract leading into the parotid tissue near the facial nerve, the tract was completely excised. Facial nerve was found clinically intact post operatively.

We suggest that patient must be subjected to all the possible investigations and imaging regarding the status of parotid gland and its relation to facial nerve and subsequent Parotidectomy if required.

Since majority of these lesions are subtle in presentation the surgeon more often than not are tempted to do a local excision which very often leads to recurrence the incidence of which is as high as 50%. In the series of Ford etal the mean number of operations before permanent cure was 2.4%. Permanent cure can only be achieved after complete excision of these lesions which involves Parotidectomy and exposure of the facial nerve (required in 92% cases of Triglia J M etal) [3] major risk of surgery being facial nerve palsy. Embryologically the defect is superficial to the facial nerve

but practically the relationship between the two is highly variable with 30% cases the tract passes medial to the facial nerve. More caution is needed in management of the Cysts as they are always located above the main tract of the facial nerve. Care must be taken in removing a small segment of the cartilage of the External ear canal in the terminal part of the tract. Occasionally these lesions are difficult to diagnosis especially in sinus or fistula cases without external opening. They must be suspected in cases of Otorrhoea without Otitis media with am orifice communicating with the External auditory canal; in patients with an opening located in the neck in an area bounded Sternocleidomastoid muscle behind, hyoid below, and the angle of the mandible in front, in case of isolated parotid cyst which increases in size during attacks of inflammation [3].

Conclusion

From all Branchial cleft anomalies, the most common defect arises from the second Branchial arch, which makes up almost 95% of defects, then the first arch of less than 6% of defects. These lesions are difficult to diagnose clinically and a high degree of suspicion is needed in diagnosis. Patient must be subjected to all the possible investigations and imaging regarding the status of parotid gland and its relation to facial nerve and consent of Parotidectomy must be taken before surgery. Results are good if preoperative evaluation is good and tract removal is complete including removing a small segment of the cartilage of the External ear canal in the terminal part of the tract.

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