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A Case Report of an Unusual Collateral Fistula

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Authors' contributions

This work was carried out in collaboration between both authors. Author JV designed and wrote the first draft of the manuscript. Author MNK managed the literature searches and approved the final manuscript.

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Case Report

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ABSTRACT

This is a case report of a 20 year old female who presented with recurrent pre auricular swelling and ear discharge since childhood. Upon further investigation, she was found to have left superficial preauricular fistula communicating with posterior auricle with no communication with internal or external auditory meatus. Cervico-aural fistula is a rare variant which accounts for less than 8% of all branchial arch abnormality [1].

Keywords: Collaural fistula; 1st branchial arch anomaly; superficial fistula.

1. INTRODUCTION

In view of rare variant, the average age of presentation of 1st branchial arch anomaly is 19 years and commonly associated with diagnostic dilemma which leads to an average delay of 3.5 years from age of presentation to correct diagnosis [2]. Recurrent otorrhea in the absence of chronic otitis should raise the index of suspicious towards 1st branchial arch anomaly. However only 44% of patients present with sinus/fistula opening at EAC [3]. 1st branchial arch abnormality can present as accessory tragi, preauricular sinuses, cyst or colloaural

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fistula. It usually communicates from tragal notch down to neck or external auditory meatus and opens in-between angle of mandible and sternocleidomastoid muscle. In exceptional cases, the fistula can communicate medially to Eustachian tube opening or internal and external carotid artery.

The fistula is more commonly seen on the left side and twice more common among women compared to men [2]. As reported in this case the women presented with left sided 1st branchial arch anomaly.

2. CASE REPORT

20 year old female presented to our centre with chief complain of recurrent left ear discharge since childhood which was never investigated before. She brought it to medical attention this year because for the past one year there has been more *frequent ear discharge* and recurrent swelling at preauricular region. No other associated symptoms such as otalgia, otorrhea, hearing loss, vertigo, tinnitus, trauma or neck swelling.

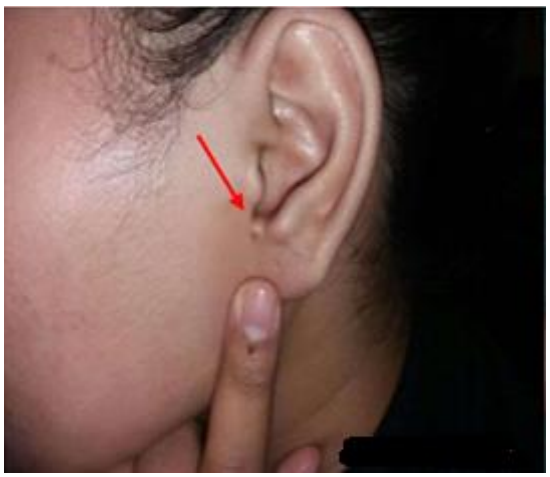

Fig. 1. Shows the pin hole size opening inferior to intratragal notch

On examination there was pin size opening inferior to intratragal notch with crusting at left posterior auricular region behind ear lobe. Otherwise no swelling or discharge from the opening.

During the initial visit she was given oral amoxicillin and planned for fistulogram as an outpatient. Fistulogram revealed that she has a superficial left subcutaneous preauricular fistula communicating with left posterior auricle. The tract measured 1.5 cm in length with no extension into internal or external acoustic meatus. Ossicles and middle ear structures were intact. No lytic sclerotic bone lesion and mastoid air cells were clear.

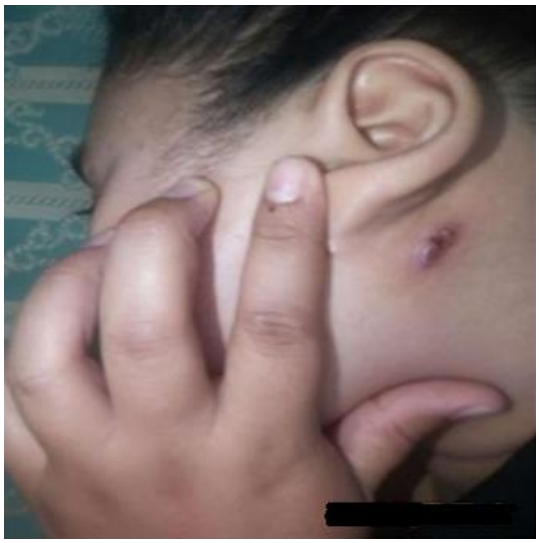

Fig. 2. Crusting at postauricular region noticed during examination

She underwent left preauricular fistula excision. Intraoperatively, there was left preauricular fistula tract extending posteriorly to post auricular region measuring about 1.5 cm and the whole tract was completely excised. Facial nerve or parotid gland was not encountered in view the tract was superficial.

The present study review at clinic postoperatively, the surgical scar healed well and no more discharge. The fistulous tract was completely excised.

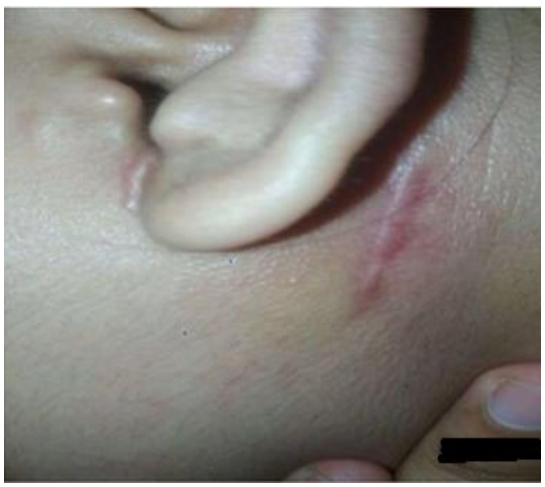

Fig. 3. Post operative scar

3. DISCUSSION

Buccopharyngeal membrane, which separates mouth from the pharynx, disappears by the end of the third week after forming bars at the pharyngeal walls. The mesodermal condensation of these bars is known as branchial arches. The ventrally fused u- shaped arches form the support of pharynx and it consists of six arches whereby the fifth arch becomes rudimentary. Branchial cleft forms arch laterally and pouch internally.

Derivatives of 1st branchial arch are mandible, maxilla, sphenomandibular ligament, incus, malleus, anterior malleus ligament and muscle of mastication. External auditory meatus (EAC) arises from the groove and eustachian tube and middle ear from the pouch of 1st branchial cleft. 1st branchial cleft is the only cleft that does not obliterate at the end of embryonic development [3]. Only the dorsal part of 1st groove takes part in forming EAC and the ventral groove usually disappears. However, in rare occasion it can persist and present as preauricular sinus or collateral fistula later in life as reported in the above case.

Arnot (1971) classified 1st branchial cleft anomaly to type 1 and type 2 based on anatomical variant followed work in 1972 who took both histology and anatomy into consideration and came up with more detailed classification. In 1980 Olsen et al. further classified the anomaly to cysts, sinuses or fistulas of lobule, canal, post auricular region or angle of jaw. Point of time at which the disorder develops determines whether it's Type 1 or Type 2 anomaly.

Comparison between Arnot and Work Classification on 1st branchial cleft anomaly:

	Arnot	Work
Type 1	Cyst or sinus ectodermal in origin at parotid gland	Lesion is adjacent to EACmedial to conchae and extends to postauricular crease or supra auricular region. Its ectodermal in origin and usually lies superior to main trunk of facial nerve.
Type 2	Sinus or fistulous tract opens between upper neck and EAC	Cyst, fistula or sinus is usually both ectodermal and mesodermal in origin arising between upper neck and angle of mandible. The lesion passes through parotid gland close to facial nerve in variable relation. The fistula usually opens below angle of mandible.

Various presentation has been reported for type 2 first branchial arch anomaly which includes chronic purulent ear discharge with sinus openings at floor of EAC, fistula or abscess discharging in Poncet's triangle at lateral aspect of neck above hyoid bone and anterior to sternocleidomastoid muscle [4,5], (the position of sinus or fistula opening at neck varies from pre, infra or post auricle to submandibular region), preauricular cyst, swelling at parotid region or fistulous opening anterior to fossa of rosenmuller at lateral nasopharyngeal wall which is an extremely rare [6].

Our case is classified as work's Type 1 based on the location of fistulous tract which was medial to ear conchae and the tract was extending posteriorly to post auricular region.

Obliteration of ventral part of 1st branchial arch takes place simultaneously with emergence of parotid gland as ectodermal ingrowth and migration of facial nerve from second branchial arch which explains the close relation between 1st branchial cleft anomaly with parotid gland and facial nerve [4]. In 2003 Solares, et al. reported on relation of facial nerve to 1st branchial cleft anomaly in 10 patients whereby 2 of the lesion ran lateral to facial nerve, 7 ran medial and only 1 ran in between branches of the nerve [3]. In general sinus tract runs superficial to facial nerve compare to fistulous tract and in presence of opening at EAC the tract mostly related superficially to facial nerve [7,8]. However, facial nerve was not encountered in this case report during surgery because the fistulous tract was superficial and according to Work's Type 1 classification, the tract usually runs superior to main trunk of facial nerve.

In addition to history and clinical presentation, radiographic visualization is important to establish diagnosis and in planning for surgery. In a case report by Jain, et al. published in 2014 they found CT sialography to be more superior diagnostic tool compare to CT fistulography [9,10] and suggested CT sialography as gold standard imaging tool to diagnose pre auricular sinuses or fistulas [11]. Due to unavailability of CT sialography, we used CT fistulography as imaging tool and complete surgical excision was still feasible. From our opinion, both CT sialography and fistulography can be useful measures. The cost and availability of facility in particular centre needs to be taken into consideration before choosing imaging modality.

Complete surgical obliteration of sinus or fistulous tract is the mainstay and definitive treatment however it should only be carried out after eradication of infection [5]. Prior to surgical

exploration facial nerve will need early identification and proper exposure as precaution since there is high risk of injuring the nerve leading to post operative facial nerve palsy. As an alternative, facial nerve monitor can also be used [12].

4. CONCLUSION

In conclusion, 1st branchial arch anomaly is a challenging diagnosis to make in view of presents of multiple variant and it is usually not associated with other facial malformation. In younger patients presenting with recurrent ear or neck discharge a detailed history, otology and neck examination would aid in achieving accurate diagnosis earlier. Complete excision of the lesion after infection has settled is the mainstay of treatment and attempting surgical drainage may complicate definitive surgery.

CONSENT AND ETHICAL APPROVAL

As per university standard guideline patient's consent and ethical approval has been collected and preserved by the authors.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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