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Collaural Fistula: A Case Report

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ABSTRACT

Cervico-aural [collaural fistula] fistula is rare and it accounts for less than 5% of branchial cleft anomalies. In this paper, we report one such case of a 9 year old girl who was presented to us with two discharging cutaneous openings on the right side; one in the floor of the external auditory canal and another in the neck at the junction of the upper 2/3rd and lower third of the sternomastoid muscle along its anterior border.

Keywords: Collaural fistula, branchial cleft, cutaneous opening

INTRODUCTION

Embryological anomalies of the first branchial cleft usually present as cysts, swellings, or fistulas in the pre-auricular or post-auricular area or high in the neck, which may become infected^[1]. Failure to recognize these unusual cases may result in misdiagnosis, inadequate treatment, and subsequent recurrence. Further definitive surgery may thus be complicated^[1].

During the 4th week of human embryological development, 6 pairs of branchial arches which will form the future lower face and neck^[2] appear, and these will disappear by the 7th week. Mesodermal in origin, these arches are separated from each other by the five branchial clefts externally and five pharyngeal pouches internally. First branchial cleft anomalies are the results of incomplete closure of the cleft.

In 1971, Arnot^[3] anatomically classified the defects as Types 1 and 2. Type 2 defects appear in the anterior cervical triangle, with a communicating tract to the external auditory canal and these usually develop during childhood.

In 1972, Work^[4] proposed a histological classification based on the presence or absence of mesothelial elements with in the wall. Type 1 lesions are of ectodermal in origin and are present medial to the concha. Meanwhile, Type 2 lesions are both ectodermal and mesodermal in origin and contain mesodermal structures such as cartilage and hair follicles. The lower opening of Type 2 fistula is usually below the angle of mandible.

CASE REPORT

A 9 year old girl, who was presented to us, had two cutaneous openings; one in the right side of the neck i.e. at the junction of the upper $2/3^{rd}$ and lower 3^{rd} of the anterior border of sternocleidomastoid muscle and the upper

cutaneous opening in the floor of the external auditory canal in the cartilaginous portion. A scar of the previous surgery performed 3 years back could be seen present over the lobule of pinna, suggesting a prior failed surgical manipulation.

At presentation, there was serous discharge from both the openings. Methylene blue injected from the cervical opening was seen coming out of the opening from an area near the scar of the previous surgery, i.e. on the lobule of the ear, which clinically confirmed the diagnosis of collaural fistula. An MR scanning in Figure 1 may show the track; however, its relationship to the facial nerve will not be demonstrated.



Figure 1.

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Surgical excision of these lesions is usually advocated. This necessitates the dissection of the track in its entirety. In most cases, the fistula runs deep to the branches of facial nerve, which may be adherent if there has been recurrent infection, though the relationship is inconstant.

Surgical excision was carried out by first keeping a horizontal skin crease incision, including an ellipse surrounding the cutaneous neck opening. Dissection was done along the subcutaneous plane, with the track being carefully separated from the surrounding tissue. Methylene blue, which was injected prior to surgery, is a useful guide to trace the tract (see Figure 2). Meanwhile, a second horizontal skin incision was placed just postero inferior to the angle of the mandible to trace the tract further upwards. It was found that the tract was passing immediately lateral to the trunk of the facial nerve just before it entered the parotid gland. It was carefully separated from the facial nerve and hence, nerve preservation was accomplished. On tracing it higher, the fistulous track was found to be getting attached to the floor of the external auditory canal near the lobule. The fistula was excised in toto.



Figure 2.

DISCUSSION

First branchial cleft anomalies are uncommon and they comprise less than 5% of all branchial cleft anomalies^[1]. Although congenital in origin, first branchial cleft anomalies can present later in life. Otorrhoea is the most common otological symptom and the condition should be suspected if recurrent otorrhoea is present in the absence of chronic otitis.

Clinically, they may present with repeated episodes of infection of the lesion. This may manifest itself with a cystic swelling or discharge from a fistulous opening either pre-auricularly or post-auricularly in the cheek, or high in the neck. A thorough otological examination should be performed in all cases as this may reveal a pit visible in the external canal at the site of entrance of a sinus or a fistula. Such a lesion may result in otorrhoea or otitis externa with infective exacerbations. A sinus/fistula opening in the external auditory canal is present in only 44% of patients, and even if such an opening is present, it may not be obvious^[2]. Sinuses and fistulae arise from incomplete closure of the first branchial groove and if the failure occurs during the time of formation of the closing membrane, a fistula extending into the middle ear may develop.

There may however be a complete absence of signs in the external auditory canal. The patient may also give a history of having to repeatedly undergo incision and drainage of an apparent abscess around the ear because of the infective exacerbations of the lesion that has not resolved. First branchial cleft anomalies can also be associated with other otological conditions.

For a Type 2 lesion, an early identification of the facial nerve at the stylomastoid foramen is recommended. If this part is affected by a disease, identifying the facial nerve proximally in the temporal bone and tracing it distally are probably the safest option. The relationship of the lesion to the facial nerve is variable. In a series of 10 patients with first branchial cleft anomalies reported by Solares $et\ al.^{[2]}$, 7 lesions ran medial to the facial nerve, 2 were lateral and 1 ran in between the branches of the facial nerve. Fistula has the tendency to run deep into the nerve, whereas sinus tracts tend to run laterally to it. Due to its variable relationship with the nerve, its removal warrants an early identification and a wide exposure of the nerve^[5], and/or the use of facial nerve monitoring.

An accurate diagnosis of first branchial cysts located in and around the parotid gland can be difficult without any surgical exploration^[5]. Poncet's triangle is the anatomical triangle, where first branchial cleft cysts or their sinus orifice are typically located. The limits of the triangle are the external auditory canal above the mental region anteriorly and the hyoid bone inferiorly.

Should the sinus/fistula opening involve the external auditory canal, it is removed with skin and cartilage. A primary closure is normally possible but if more than 30% of the circumference of the external auditory canal is denuded, split-thickness skin grafting and stenting are recommended. If the tympanic membrane or middle ear structures are involved, a reconstructive otologic surgery may be necessary.

Surgical exploration and excision is the definitive treatment of these defects and this should only be undertaken when any acute infective episode has cleared^[4]. An acute infection makes dissection more difficult and also increases the risk of injury to the surrounding structures. The potential postoperative complications are facial nerve paralysis and recurrence of the lesion. Moreover, recurrent cutaneous infections and aural discharge can occur in patients with first branchial cleft fistula if no definitive surgical intervention is done.

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CONCLUSION

In conclusion, cysts, sinuses, or fistulous tracts, which are pre-auricular or post-auricular or high in the neck, may represent a branchial cleft anomaly. A high index of suspicion is required, while a thorough otological examination and careful history should be taken into consideration. If suspected, patients should be referred for a specialist otological opinion as attempted drainage may further complicate definitive surgery. It is important to note that a conventional operation for first branchial cleft fistula is highly complicated. One of the most frequent complications is facial nerve palsy because of the close relationship between the fistulous tract and the facial nerve.

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