Archives of Orofacial Sciences
www.dental.usm.my/aos/ © Penerbit Universiti Sains Malaysia. 2018
print ISSN: 1823-8602
e-ISSN: 2231-7163

Arch Orofac Sci (2018), **13**(1): 50-53.

Case Report

Second branchial arch fistula masqueraded as recurrent parotid abscess: a case report

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Submitted: 12/12/2017. Accepted: 03/06/2018. Published online: 03/06/2018.

Abstract Branchial apparatus anomalies usually manifest in teenage or early adult life. Infection complicates second branchial pouch anomalies usually presented as a neck lump or discharging sinus. It is the most common form of anomalies compared to another branchial pouch aberrant. However, it is extremely rare to find a complete branchial fistula with both internal and external openings. Misdiagnosis usually occurs leading to inappropriate and suboptimal treatment. Here, we report of a case of complete second branchial pouch fistula and discuss the clinical presentation and surgical management of such lesion.

Keywords: Complete branchial fistula; drainage; neck; parotid abscess; second branchial arch.

Introduction

Branchial anomalies compose approximately 30% of congenital neck mass and present as cyst, sinus or fistula (Acierno and Waldhausen, 2007). Cysts are remnant of the cervical sinus without an external opening. Sinuses are persistence of the cervical sinus with external opening, whereas fistula also involves persistence of the branchial groove with breakdown of the branchial membrane resulting in a pharyngocutaneous fistula.

Second branchial cleft anomalies are the most common branchial cleft malformations (Singh *et al.*, 2012). However complete branchial fistulas are extremely uncommon with only very few reported cases. There is slight female preponderance and more common on the right side (Shankar *et al.*, 2012). Imaging is the cornerstone investigation in establishing the correct diagnosis by demonstrating the patency of the fistula. The most accepted modality of treatment is complete excision of fistulous tract by combining transoral and transcervical approach (Proctor and Proctor, 1970).

Case report

A 14-year-old female was referred with unresolving recurrent right parotid abscess since childhood. She presented with recurrent right infra-auricular swelling since the 6 years of age, which would either self-ruptured or drained with significant amount of pus. Symptoms were also associated with thick foul-smelling secretion intra-orally.

On physical examination, there was right infra-auricular swelling measuring 1 cm x 1 cm, which was non-tender with severely scarred overlying skin and keloid. There was no obvious opening surrounding the lesion. Oral hygiene examination was fair with no pus from the opening of right Stensen's duct upon milking of the parotid gland. Oropharynx was unremarkable with no pus or sinus seen at anterior pillar.

Clinical diagnosis of branchial arch fistula was made. Computed tomography (CT) scan showed a well-defined tract from right parotid to right tonsillar region (Fig. 1), which confirmed the diagnosis.

The patient was taken for excision of fistula, combining transparotid and

intraoral approach, under general anaesthesia. Tonsillectomy was performed, and the internal opening of the fistula identified at right tonsillar fossa. Elliptical incision made around the skin defect and external opening was identified and tracked until tonsillar fossa using methylene blue dye and metal probe. Blunt dissection was carried out to carefully separate the tract from the surrounding soft tissue; specially to avoid from injuring important structures in the region especially facial nerve. Facial nerve monitoring (stimulator and monitor) was also used to ensure the safety of the nerve.

Operative findings revealed that the fistula started laterally from antero-inferior to tragal cartilage, as seen after the scarred tissue was removed (Fig. 2). Then it went posterior and medial to the right parotid gland to reach the right tonsillar fossa. The fistula was successfully removed in total. The length of fistula was 5 cm from skin to right tonsillar bed (Fig. 3). Oropharyngeal defect was closed with Vicryl 3/0. Tissue glue was used to fill up the cavity and the skin was stitched in layers. Histopathological report confirmed that the specimen sent was consistent with fistula tract.

Discussion

The embryonic differentiation of branchial apparatus occurs between the 3rd and 7th week of intrauterine life. Failure of the arch tract to obliterate would result in the formation of a branchial sinus and fistula (Shankar *et al.*, 2012). The second branchial arch's sinus and fistula present in the neonatal period with an external opening along the line joining the tragus and the sternoclavicular joint along the anterior border of sternocleidomastoid muscle (Shekhar *et al.*, 2005). However, the external opening commonly situated at the lower, anterolateral neck (Acierno and Waldhausen, 2007). To the best of our knowledge, we could not find any other case reporting the opening higher up as in the present case. The second pouch gives rise to the tonsillar and supratonsillar fossa, which explains why the second cleft anomalies had entered the supratonsillar fossa.

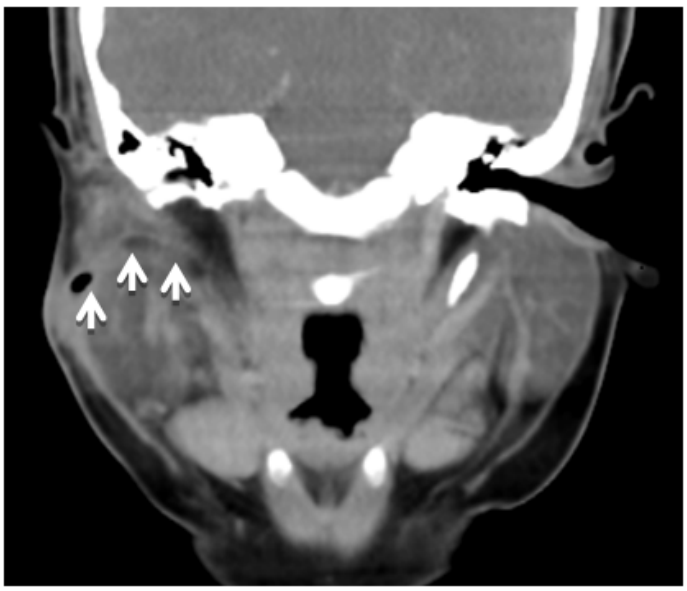

Fig. 1 CT scan showing fistulous tract superior to the right parotid to the tonsillar region (arrowheads).

Fig. 2 Fistula tracked from external opening until tonsilar fossa using metal probe.

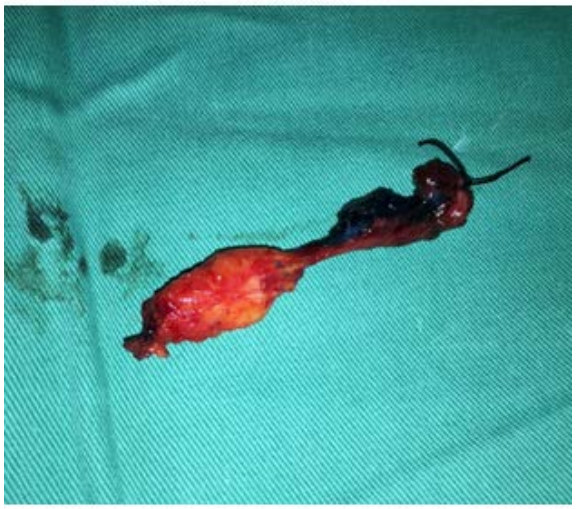

Fig. 3 Dissected specimen measuring 5 cm from skin to right tonsillar fossa.

The patient usually suffers from recurrent mucopurulent discharge from an external opening in the anterior triangle of the neck along the anterior border of the sternocleidomastoid between its middle and lower thirds (Al-Ani and Al-Fehadawi, 2015). The challenge in our case was that, the recurrent infected swelling was at right infra auricular region, not the typical presentation of branchial fistula opening. In addition, recurrent infections, multiple incision and drainage lead to the scarring of the overlying skin making the external opening no longer visible. These factors had caused the misdiagnosis of parotid abscess that was treated prior to referral, leading to delay in the definitive management of brachial fistula. During surgery, the opening was found hidden by the distorted, repeatedly incised skin forming the scarred tissue.

Imaging is essential to demonstrate the patency of the fistula, which would lead to the correct diagnosis. Preoperative imaging of tract with contrast material demonstrate the entire course of the tract, aid into surgical planning, differentiate between sinus and fistula, and minimise the chance of recurrence (Proctor and Proctor, 1970). The fistulography is a cost-effective method of showing the exact anatomy of these fistulae tract and is most often the commonest investigation available (Bist *et al.*, 2016). CT scan and magnetic resonance imaging (MRI) of the neck are helpful to produce cross sectional images of the organ and other internal body structures, useful in outlining the relationship of surrounding neurovascular structures to the lesion (Bist *et al.*, 2016). In our case, CT scan revealed a well-defined tract from right parotid to right tonsillar region which confirmed the diagnosis of branchial fistula.

The definitive treatment of branchial anomalies is complete surgical excision. A careful exploration for fistula tract must be performed with a complete excision of the entire tract. Cannulating the tract with a 2-0 or 3-0 monofilament suture or probe can facilitate fistula excision. The tract can also be injected with methylene blue; however, this may stain the surrounding tissues making dissection difficult (Acierno and Waldhausen, 2007).

Complete excision of the fistula is difficult with external approach alone. Recurrence rate of 3% has been reported with open approach alone of fresh cases and up to 20% of second surgical attempts (Bist *et al.*, 2016). This most probably due to incomplete surgical excision of the fistula tract in the parapharyngeal space (Ford *et al.*, 1992). Incision and drainage of the initial infected fistula also increase the risk of recurrence. In complete branchial fistula with a probe *in situ*, the external approach can be combined with intraoral route. No recurrences have been documented with this combined approach (Shankar *et al.*, 2012).

In this patient, we opted to excise the fistula using external approach combined with intraoral route. This technique ensured complete excision of the fistula tract and thus eliminates the chances of recurrence.

Conclusion

The present rare case highlights the importance of suspicion of other lesion than just a simple abscess, especially in a case of recurring lesion at the same site. Branchial arch anomalies must be suspected as it is common to prevent with cutaneous openings either in sinus or fistula. In a not resolving lesion such as in this case, a radiological study is warranted and referral to the appropriate faculty is warranted for the ultimate solution.

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