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Case Report

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First Branchial Cleft Cyst Type II

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Abstract

Branchial cleft defects are interestingly rare and so are often not considered as a differential diagnosis. The following is an incident of the anomalie in a 20 year old Sudanese female with a swelling that was misdiagnosed and hence not treated adequately. The swelling started 2 years ago on the right parotid area with no neurological manifestations of facial nerve injury, with a cystic content that ruptured leaving a fistula behind. After MRI was done the fistula was determined, surgical excision of both the swelling and fistula was done. On follow up, no recurrence was noted.

Introduction

Fortunately, anomalies are rare. The diagnosis and investigations are often cubersome. Having said that, patients often go through a lot of investigations and at times, invasive procedures, until the correct diagnosis is made, making treatment challenging. Hopefully, with more reports these anomalies are better understood.

They were first described by Work and Proctor in 1963 as two types [1]. It makes less than 8%-10% of all branchial arch cleft defects [2,3]. Somashekara, et al. stated that this anomalie makes less than 1% of all branchial anomalies, that is approximately 200 cases [4].

Type I defects are those of ectodermal origin, and clinically present as a duplication of the membranous external auditory canal. Type II, is of both, ectodermal and mesodermal origins, hence, a duplication of both cartilaginous and membranous external auditory canal [1,5]. Volaris and Pahor introduced a third variant, type III, where in the fistula tract extending from the ear canal to the mastoid and sternocleidomastoid muscle [2].

In order to understand this classification, it's essential to have a solid understanding of embryology. In brief, this anomalie is likely to occur during the 4th-5th week interval of gestation. The foetus has five mesodermal arches, four ectodermal clefts and four endodermal pouches. The concern of this anomalie is in the first branchial pouch and cleft, from which the Eustachian tube and tympanic cavity, and the external auditory meatus and part of the concha emerge, respectively [1,5,6].

This is a case of Type II Work classification of a cystic fistulated swelling.

Case Report

A 20 year old Sudanese young lady presented with a 2 year old soft painless swelling in the parotid area on the right side that gradually increased in size. A general surgeon attempted to drain the contents of the swelling that was mistaken for an abscess. The wound did not heal and kept discharging for over 5 months.

The patient mentioned having experienced a chest infection right before these events escalated. Otherwise, she had no medical history of significance and denied habits such as snuff dipping or alcohol consumption.

On examination, the swelling was found to be non-tender, fluctuant, rubbery, mobile, lobulated of a size 5x4 cm, with a horizontal scar halfing it into two, extending from the angle of the mandible and into the area of the tail of the parotid anterioposteriorly, raising the lobe of the ear. The skin was intact and of normal colour (except for the scar) without apparent discharge (Figure 1). The facial nerve was intact and symmetrical.

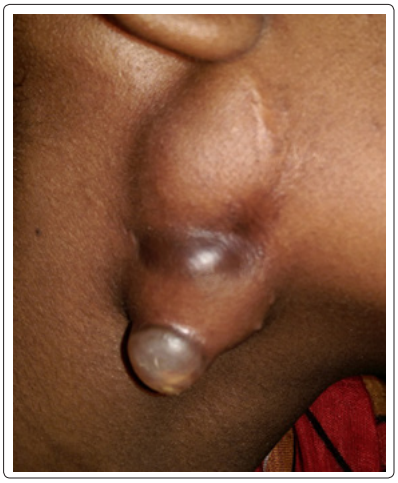

Figure 1: Shows mobile rubbery and soft fluctuant lobulated swelling in the tail of the parotid area raising the ear lobe and extending to the angle of the mandible with the scar of the previous operation

Intraorally, there was no apparent soft palate bulging or deviation of the uvula or paratonsillar pillars or fossa, excluding the involvement of the deep lobe or parapharyngeal space.

After a week of the patient's first presentation, the swelling ruptured creating a fistula (Figure 2). FNA was requested but was inconclusive, revealing inflammatory and epithelial cells. Hence, and MRI was done (Figure 3). The report revealed a well-defined, hyper intense, homogenous, parotid lobulated mass with incorporating of a fistulous tract at the tail of the parotid, external auditory meatus and soft tissue overlying the angle of the mandible.

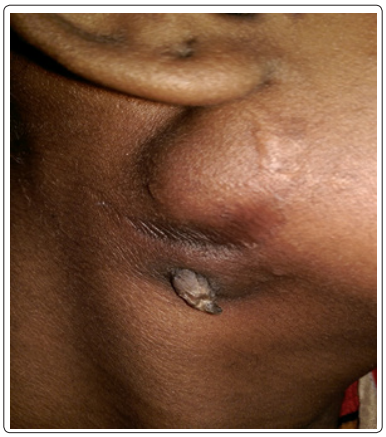

Figure 2: Shows rupture of the flucuant part of the swelling and a fistula at the angle of the mandible after a week of the diagnosis

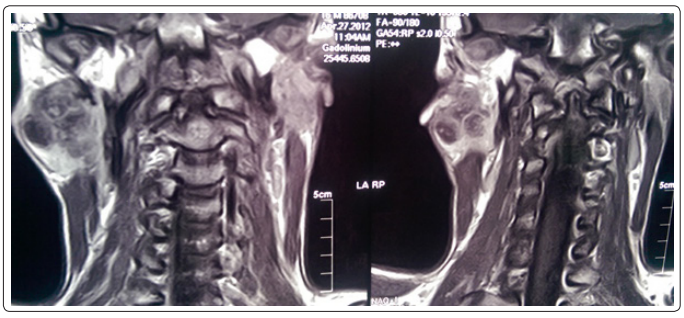

Figure 3: Magnetic resonance imaging showing a well-defined hyper-intense homogenous parotid lobulated mass with incorporation of a fistulous tract at the parotid tail, external auditory meatus and soft tissue overlying angle of the mandible

Partial superficial parotidectomy using Modified Blair's incision with excision of fistula and affected skin along with affected cartilage was decided and done successfully (Figure 4). The facial nerve was dissected, isolated and preserved.

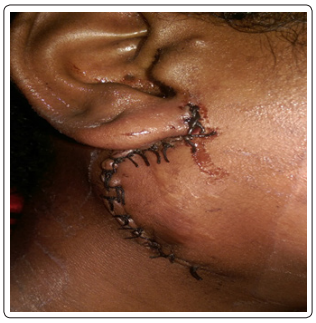

Figure 4: Shows modified Blair incision of the partial superficial parotidectomy with excision of the affected skin and fistulous tract

Discussion

Triglia, et al. postulated that the fistula has been found to pass deep to the nerve in 28%, 8% split around the nerve, and 64% were superficial [5,7].

When the sinus of His fails to obliterate it causes branchial anomalies [4,8]. In foetal development, the clefts obliterate ventro-dorsal, therefore, anomalies occurring closer to the ear and parotid - dorsally - occur more commonly than in hyoid region - ventrally [9]. This area or triangle is known as Poncet's triangle (boundaries: superiorly external auditory canal, anteriorly mental area, inferiorly hyoid bone) [4]. The presentation of this case was consistent with type II.

According to the literature, the regions most affected are cervical and parotid, twice as common in women than men and fistulas twice as much on the left side than right side [6]. All the above is consistent with our case except for the side, the lesion is on the right side. Regarding the site, this explains the Poncet's triangle, and any lesion in this triangle with such a history of benign purulent swelling, a differential of FPBC anomalie should be considered.

The patient mentioned a chest infection that preceded the swelling. Codreanu CM, et al. reported acute rhinopharygintis after which the swelling became reddish and started to discharge whitish fluid [10].

Regarding management, Work suggested marsupelization for type I through external auditory canal and excision with superficial paritodectomy for type II using modified Blair's incision or S-incision, which was implemented for our patient [2]. Sometimes the external auditory meatus is included if it as involved [2]. Of course, great care should be taken to identify, dissect and protect the facial nerve, to avoid palsies [3]. Tracing the fistulous tract is thus essential and can be achieved with CT fitulogram with the help of dyes such as diatrizoate meglumine (renografin) infusion or MRI as was done with our patient [6]. In a study of 39 cases, only 3 came back with a recurrence, seeking a second surgical treatment [7,11]. Our patient was followed up with no recurrence or other complains to date.

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