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Case Report

A First Branchial Cleft Cyst Masquerading as a Chronic Non-Healing Wound

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ABSTRACT

Background: First branchial cleft anomaly exhibits variable clinical features, ranging from a painless swelling, discharging sinus or pit, to recurrent infection. It could be easily misdiagnosed and mismanaged, resulting in recurrence. Hence, any swellings or pits in Poncet's triangle with a history of recurrent infection should raise the suspicion of a first branchial cleft anomaly. **Objective:** To present a case of chronic non-healing wound caused by an incompletely excised first branchial cleft anomaly. **Methods:** Case observation of a patient with a history of incomplete excision of a first branchial cyst. **Results:** Our patient was a case of incompletely excised first branchial cyst which subsequently presented as a chronic non- healing wound. She had undergone several workup for non-healing wound resulting in delay in receiving definitive treatment. **Conclusion:** Early recognition of first branchial cleft anomalies is important to prevent chronic complications and mismanagement.

Keywords: branchial cleft cyst, chronic wound, wound healing

1. Introduction

The first branchial cleft anomaly is a rare developmental defect, typically presenting as a cyst, sinus, or fistula near the ear, in the parotid or upper neck regions. It is frequently accompanied by concurrent infections, which could be misdiagnosed and mismanaged, resulting in recurrence. The delay between the first presentation and appropriate management can span up to 3.5 years [1]. This case report highlights the occurrence of an incompletely excised first branchial cleft cyst that presented as a chronic, non-healing wound, emphasising the importance of maintaining a high index of clinical suspicion for an accurate diagnosis at the time of first presentation.

2. Case Presentation

A 21-year-old Malaysian female presented with a painless, slowly-enlarging swelling in the right upper neck swelling of 1-year duration. The swelling was not associated with fever, otalgia, otorrhea, or skin changes. Clinical examination revealed a 5x2cm soft, mobile, non-tender swelling at right neck level 2. The examination of ear and oral cavity were unremarkable. There was no preauricular skin tags or pits. Neck ultrasonography identified a cystic lesion measuring 1x2x6cm in the right infraauricular region. It initially diagnosed as a right infraauricular epidermal cyst which was excised without intraoperative complications. The wound healed well post-operatively, and histopathological examination confirmed the diagnosis of an epidermal cyst.

However, two months later, the patient complained of recurrence of swelling which became infected and required an incision and drainage. 3cc pus was drained. The wound failed to heal completely and had persistent mucopurulent discharge (Fig. 1). Histopathology of wound edges revealed inflamed granulation tissue without signs of tuberculosis, fungal infection or malignancy. A computed tomography (CT) fistulogram was performed to investigate the possibility of a fistula tract, which revealed a well-defined hypodense lesion at the subcutaneous layer of the right infraauricular region with a thin enhancing wall but no fistula tract was demonstrated. The lesion did not show a clear plane with the right parotid gland (Fig. 2). During wound exploration of the right neck wound, a sinus tract was found superficial to the right parotid gland, extending superiorly, and attaching to the right tragal perichondrium (Fig. 3). Sebaceous material was draining from the tract. The sinus tract, along with a small cuff of tragal perichondrium, was excised, and the wound was closed primarily with successful postoperative healing. The superior end of the tract was cut open and sebaceous material was demonstrated (Fig. 4). Histopathological examination of the excised tissue confirmed the presence of a sinus tract lined by stratified squamous epithelium and in continuity with the overlying skin epidermis without evidence of granuloma, dysplasia, or malignancy.

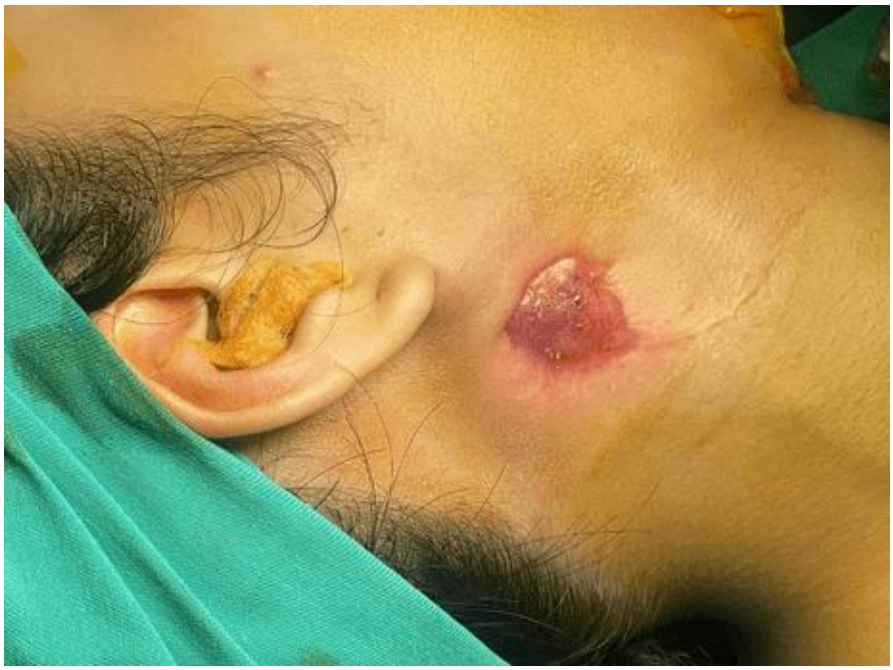

Figure 1. An epithelised wound at right infra auricular region with sloping edges and granulation tissue at base measuring 2.5cm x 2cm in continuity with well healed scar from previous excision.

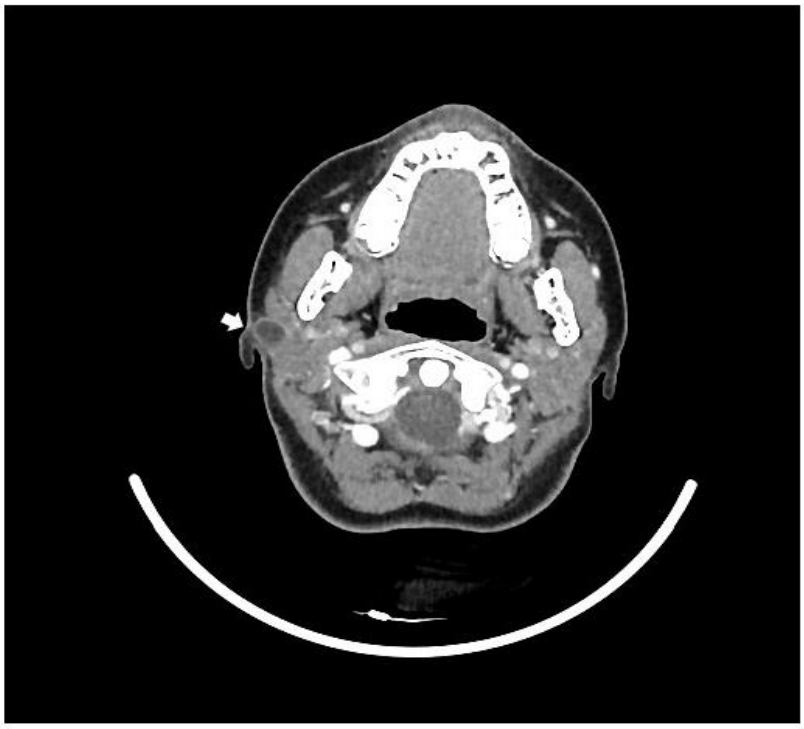

Figure 2. Axial view of contrast enhanced CT Neck showed a rim enhancing hypodense lesion (white arrow) at right postero- inferior auricular region measuring 1.3x0.8x1.4cm with fistula communication to the skin. The lesion was indenting on the superficial lobe of right parotid gland but there was no abnormal enhancement of right parotid gland.

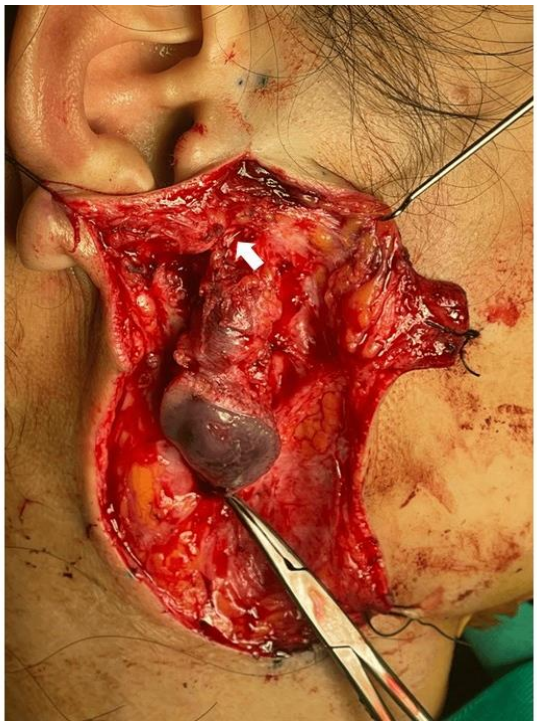

Figure 3. The wound was connected to a sinus tract leading to the tragal cartilage (white arrow).

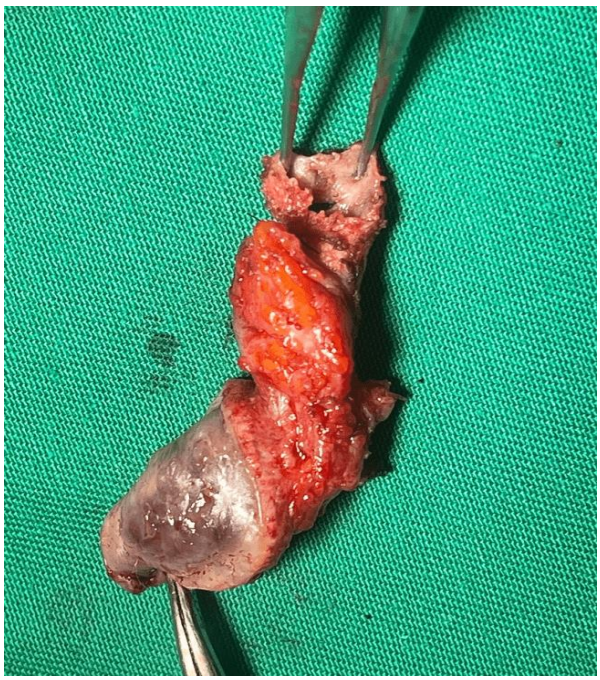

Figure 4. The wound with sinus tract and a cuff of tragal perichondrium were removed. The superior end of the tract was cut open and sebaceous material was demonstrated.

3. Discussion

The first branchial cleft anomaly is a rare condition that often lacks facial malformations, exhibiting highly variable clinical features that complicate diagnosis. The presentations can range from painless swelling, discharging sinuses or pits at the preauricular region or upper neck, to recurrent otorrhea. Recurrent otorrhea in children, particularly in the absence of middle ear disease or significant canal inflammation, should prompt the suspicion of a first branchial cleft anomaly, leading to careful inspection of the external ear canal for any pits or sinus openings [2, 3]. These anomalies are commonly located at Poncet's triangle, which is bounded by the external auditory canal superiorly, the mental region anteriorly, and the hyoid bone inferiorly [4]. Swellings or pits in this triangle, particularly with a history of recurrent infections, should raise the suspicion of a first branchial cleft anomaly. Fine needle aspiration cytology can assist in ruling out differential diagnoses such as salivary gland pathology, cystic nodal metastasis or inflammatory adenopathy [5]. While numerous classifications for first branchial clefts exist, their clinical utility is limited as they do not significantly influence the surgical planning [2].

Imaging is critical for preoperative planning. Neck ultrasonography is useful for initial assessment of a neck swelling, especially in children, it may fail to detect a thin sinus tract connecting to the branchial cyst, as seen in our patient. Contrast-enhanced CT or MRI of the neck is advocated for better delineation of the extent of the swelling and its relationship to important surrounding structures, such as facial nerve, parotid gland, external and middle ear, and the diagnosis of occult sinus tract which may be missed otherwise[6]. In cases of a branchial sinus or fistula, the cutaneous opening can be cannulated and the contrast is infused under fluoroscopic guidance. If there is prompt filling of fistula tract, additional contrast can be injected and CT fistulogram is performed for better delineation of the course of the sinus or fistula, thus, aiding in preoperative counselling and surgical planning [7]. Incomplete excision of first branchial anomalies can lead to chronic infection and recurrence, as seen in our patient, where the presence of sinus tract was not initially identified. The contrast-enhanced CT and fistulogram of our patient revealed a collection at the wound with no tract was demonstrated, possibly due to a collapsed sinus tract, resulting in delayed diagnosis.

The management of branchial malformation is complete excision with preservation of facial nerve. Skin incision may include modified Blair or post auricular approach, incorporating any sinus or fistula openings. Intraoperative facial nerve monitoring is essential, as the relationship between the first branchial cleft anomaly and the facial nerve is unpredictable, especially in cases with parotid agenesis or previous incisions and drainage [8, 9]. The complete excision of first branchial anomaly, may require resection of skin and cartilage within the external auditory canal to prevent recurrence. The defect in the external canal can be closed primarily or with a split skin graft and stenting if the defect exceeds 30% of the circumference of the external canal.

Middle ear involvement may necessitate mastoidectomy with reconstruction of ot ological structures[10]. The recurrence risk increases with the number of preoperative incisions and drainage.

Our patient underwent extensive workups, including cultures and histopathological examination to exclude chronic infection such as tuberculosis, fungal infection and malignancy. The diagnosis of a first branchial cleft cyst was made intraoperatively when careful wound exploration revealed a sinus tract connecting to the non-healing wound. This case highlights the importance of high clinical suspicion, comprehensive preoperative investigation and accurate initial diagnosis to prevent misdiagnosis and treatment failure.

4. Conclusion

This case underscores the importance of maintaining a high level of clinical suspicion when patients present with pits, swellings, or sinus openings in Poncet's triangle. Alternative diagnosis should be considered in cases of chronic, non healing wounds, after excluding chronic infections such as fungal infections, tuberculosis and malignancies. A discharging sinus may contribute to the persistent non-healing nature of the wound. Thorough preoperative imaging and an accurate diagnosis are essential for the successful treatment of the first branchial cleft anomalies.

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