

First Branchial Cleft Sinus Mimicking an Epidermoid Inclusion Cyst: A Case Report

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ABSTRACT

First Branchial Cleft Anomaly (FBCA) is an uncommon congenital anomaly that may mimic other periauricular lesions. A two-year-old male child presented with a right infra-auricular pit present at birth and eight months of intermittent painful swelling with foul-smelling whitish discharge following spontaneous rupture. An examination showed a sinus tract scar. Computed Tomography (CT) sinogram demonstrated a tract extending toward the floor of the External Auditory Canal (EAC), consistent with a Type II first branchial cleft sinus. After the infection resolved, the cyst was completely resected while preserving the facial nerve. Histopathology revealed a cyst with a keratinising stratified squamous lining, lamellated keratin, and no adnexal or mesodermal structures, confirming an Epidermoid Inclusion Cyst (EIC).

Keywords: Diagnostic dilemma, External auditory canal, Histopathology, Infra-auricular region, Paediatric neck mass

CASE REPORT

A two-year-old male child presented with a chief complaint of right infra-auricular swelling for the past eight months. The swelling was intermittent (on and off) in nature and associated with episodes of pain and discharge. The child had a congenital pit at the same site since birth, which remained asymptomatic initially. At around six months of age, it began discharging whitish, foul-smelling material and subsequently developed recurrent infections, each episode resolving temporarily with treatment, followed by recurrence of swelling.

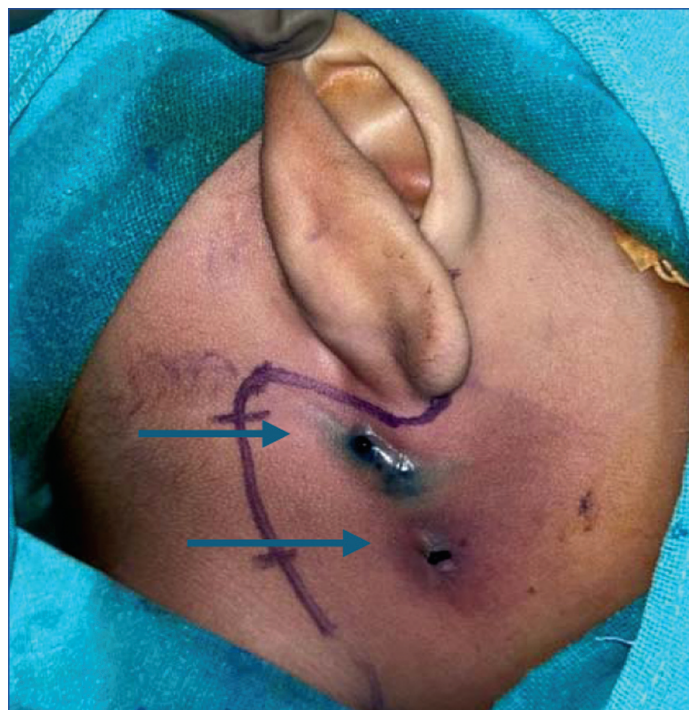
The child was born full term through normal vaginal delivery without any complications. There was a small cutaneous puncture at the right infra-auricular region that was seen right after birth, but was asymptomatic in early infancy. The swelling at the same site became painful and, approximately eight months prior to presentation, ruptured spontaneously, discharging a sticky, whitish, foul-smelling, non bloody material, accompanied by local tenderness. The child was treated at a peripheral healthcare facility with antibiotics, incision and drainage, followed by serial wound dressings. The acute infection resolved following treatment; however, a chronic healed sinus tract persisted thereafter, noted from approximately one year of age.

The child was active on admission and appeared healthy, with normal growth parameters and consistent vital signs. On examination of the right infra-auricular region, there was a healed sinus opening measuring approximately 2-3 mm, with no active discharge, erythema, induration, tenderness, or local warmth [Table/Fig-1]. The bilateral EACs and tympanic membranes were found to be normal and intact on otoscopic examination. Parotid gland swelling could not be palpated, and the functionality of the facial nerves was not impaired. General physical examination was unremarkable.

Laboratory investigations showed haemoglobin of 11.3 g/dL and a total leukocyte count of 6,400/cu mm; liver and kidney function tests, and urine analysis were within normal range.

The CT sinogram revealed a localised collection measuring approximately 1.9×1.4×2.8 cm in the right parotidomasseteric region, with a sinus tract extending toward the floor of the EAC over a length of approximately 2.5 cm, as shown in [Table/Fig-2a,b].

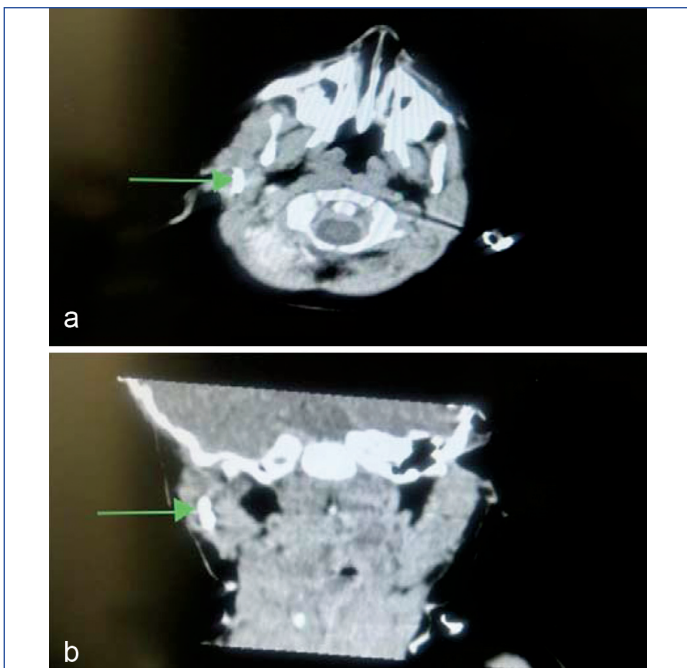
The intraoperative photograph shows the exposure of the right infra-auricular and preauricular area using a preauricular incision.



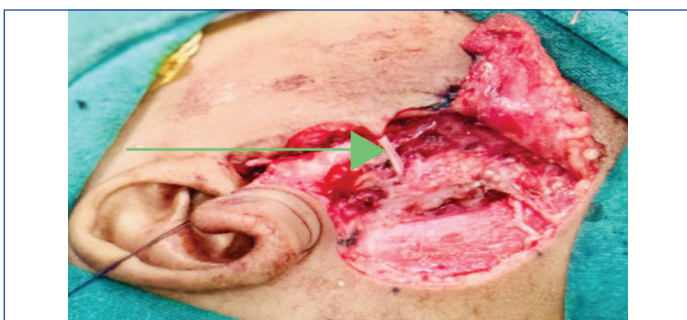
[Table/Fig-1]: Preoperative clinical photograph of a congenital pit in the right infra-auricular area (blue arrow) and with healed skin around the area as a result of previous abscesses.

The reflection of the skin and subcutaneous tissues has provided a view of the field of operation immediately adjacent to the external ear canal and ear lobe. There is a fibrous tract that is observed running parallel to the EAC, which is in line with the suspected sinus tract. There is no sign of cartilaginous attachment or middle ear communication in the surrounding soft-tissues. A close dissection was done very near the facial nerve that was detected and spared. Parotid ductal structures were not violated, and the tract was removed in totality as depicted in [Table/Fig-3].

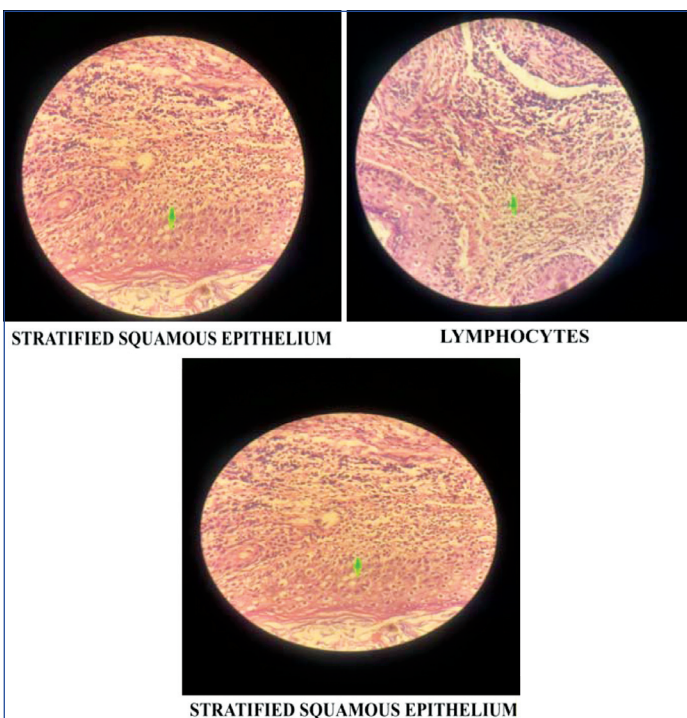
Histopathological analysis of the resected tract showed that there was a cyst that was lined by keratinising stratified squamous epithelium that had a large amount of lamellated keratin debris [Table/Fig-4]. The cyst wall was composed of fibrous stroma and a focal chronic inflammatory infiltrate. It is important to note that no adnexal structures of the skin, lymphoid tissue, or mesodermal/



[Table/Fig-2a,b]: Contrast-enhanced CT sinogram of a contrast-filled sinus tract of the right infra-auricular area. The arrow indicates the course of the sinus tract.



[Table/Fig-3]: Intraoperative image of a surgical exposure of the right infra-auricular space, through preauricular approach. The fibrous sinus tract (arrow) appears to be parallel to the External Auditory Canal (EAC) after elevation of skin flaps.



[Table/Fig-4]: Histopathology of the excised infra-auricular sinus tract (Haematoxylin and Eosin stain). a) Low-power image; a cyst wall with a keratinising stratified squamous epithelial lining (arrow); b) Second higher-power view showing dense lymphocytic infiltration in the fibrous stroma, which is characteristic of chronic inflammation; c) Another area showing stratified squamous epithelial lining that lacks adnexal structures of the skin, lymphoid aggregates and mesodermal/cartilaginous bodies, which are indicative of EICs.

cartilaginous matter were detected. Such characteristics did not fit with a branchial cleft defect. The histopathology report specifically stated that no branchial cleft sinus was present, thereby favouring the diagnosis of an EIC.

The surgical wound was seen to be well approximated with existing sutures and no signs of wound gaping, active discharge, erythema, or oedema. The ear shape was maintained, and the facial nerve was not dysfunctional [Table/Fig-5].



[Table/Fig-5]: A postoperative clinical photograph of the right infra-auricular area after the surgery of the lesion.

On postoperative day 1: The patient was stable and had no complications. Postoperative day 15: Sutures were removed, and the wound had healed well. At 6-month follow-up: No recurrence, facial nerve palsy, or residual lesion.

DISCUSSION

Congenital periauricular lesions such as Preauricular Sinuses (PAS), EICs, and FBCAs present a diagnostic challenge due to their similar embryogenesis and anatomical proximity to the EAC, the parotid gland, and the facial nerve [1,2]. These lesions often present as recurrent infra-auricular or postauricular infection and discharge, which often results in late diagnosis and inappropriate recurring interventions [3].

Work WP (1972) first proposed a widely accepted diagnostic framework for FBCA, classifying them into two types based on embryological origin. Type I lesions are purely ectodermal and represent duplications of the membranous external auditory canal, typically presenting as superficial cysts or sinuses without cartilaginous components. In contrast, Type II lesions are of ectodermal and mesodermal origin and may have deeper extensions, often associated with the parotid gland, facial nerve, and cartilaginous structures, thereby posing greater surgical complexity and risk. Importantly, Work WP emphasised that these anomalies may follow variable anatomical courses, often running parallel to the external auditory canal and in close proximity to critical neurovascular structures. However, he also acknowledged that not all lesions conform strictly to this classification, highlighting inherent limitations in applying this model to atypical or overlapping presentations [4].

This diagnostic ambiguity has been further supported by subsequent studies. Choi SJ et al., (2007) described a variant type of PAS, termed the "postauricular sinus," which accounted for approximately 10.9% of cases [5].

In a study by Dutta M et al., (2013), a total of 28 cases of epidermoid cysts were analysed. Five were female, with a male: female ratio of 4.6. The age range was two to 60 years (mean=30). Excision was the preferred treatment in 20 cases (71.4%). Various sites like the submandibular region (5), pinna (5), sublingual region (1), periorbital (6), suprasternal (6), along the anterior border of sternocleidomastoid (1) and glabella (3) were involved, along with an iatrogenic implantation epidermoid cyst in a tracheostomy scar [6].

Jain S et al., (2014) provided a vivid example of the clinical outcomes of this overlap, in which a rare Work Type II FBCA manifested as a postauricular salivary fistula. In their case, CT sialography revealed parotid duct communication, and this required superficial parotidectomy, including facial nerve dissection, and histopathology revealed ectodermal lining and salivary acini, making a true FBCA [7].

CONCLUSION(S)

The present case shows that EICs, PAS variants, and first branchial clefts exhibit significant diagnostic overlap in children with an infra-auricular sinus tract. Although clinicoradiological findings strongly suggested a Type II first-branchial cleft sinus, definitive diagnosis was established only by histopathologic analysis, which favoured an EIC.

Ethical considerations: The patient had the informed consent of his parents/guardians to be clinically evaluated, undergo surgery and have the present case report published, complete with accompanying images. The patients' anonymity has been preserved, and no personally identifiable information has been revealed. The ethical principles that guided the research are those outlined in the Declaration of Helsinki.

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