Avoiding Surgical Pitfalls during Resection of a "Hybrid" First and Second Branchial Cleft Cyst

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Introduction

- Branchial cleft cysts (BCCs) are the second most common congenital head and neck lesions diagnosed in children (1).
- Remnant tissue can persist, resulting in cysts, sinuses or fistulae, which can present as expanding neck masses, chronic neck infections, or draining sinuses that require complete surgical resection for definitive treatment.
- Second BCCs represent about 95% of branchial cleft lesions. They are often identified externally near the anterior edge of the upper third of the sternocleidomastoid muscle and travel past the internal and external carotid arteries to the tonsillar fossa (1). Histologically, they are often lined with stratified squamous epithelium or ciliated columnar epithelium.
- First BCCs, which comprise about 1% of all lesions, normally track from the external ear canal to the submandibular area, often involving the parotid gland and occasionally the facial nerve (1).
- Here we describe a rare case of a sinus tract exhibiting anatomical and histological characteristics of both first and second BCCs.

Case

- A fourteen year old female presented with a draining pit inferior to the angle of the left mandible.
- MRI with contrast revealed a tract from the skin inferolateral to the left parotid gland extending across the parapharyngeal space to the left tonsil.
- Intraoperatively, the main trunk of the facial nerve was encountered looping around the inferior aspect of the tract from posterior to anterior. The tract was excised with preservation of the facial nerve.
- Pathology revealed features of both first (fistula lined by dermal tissue) and second (cyst lined by squamous and respiratory epithelium with associated lymphoid tissue) BCCs.
- The patient remains free of recurrence with normal facial nerve function.

Operative Images

MRI

Discussion

BCCs generally present with recurrent drainage from a pit-like depression at the angle of the mandible. The internal opening of the sinus provides important information regarding the origin of a BCC (1). Complete surgical excision of the tract is critical since these tracts do not spontaneously regress and there is a high risk of recurrent infection with incomplete resection (3).

In this case, involvement of the main trunk of the facial nerve was unexpected as imaging showed a tract running inferolateral to but not involving the parotid gland. In addition, the internal opening at the left tonsil strongly supported a diagnosis of a second BCC, which typically involves hypoglossal and/or glossopharyngeal nerves (1). Finally, fistulous tracts with clear openings to both the skin and an interior space are more likely to lie deep to the facial nerve as compared with sinuses and tracts with singular openings (2). The diagnosis of a hybrid first and second BCC is supported by the pathological finding of both dermal tissue, as is found in type I first BCCs, as well as squamous and respiratory epithelium with associated lymphoid tissue, which is more characteristic of a second BCC.

Few similar cases exist in the literature. A 2008 case report documents a BCC involving the parotid gland on ultrasound (characteristic of first BCC) but with termination near the tonsillar fossa (characteristic of second BCC). The tract was found to displace the facial nerve. A second case reported in Singapore in 1991 describes a fistulous tract with features of both a first and second BCC anomaly (4).

This case exhibits anatomical and histological findings that defy the current understanding of the origins of BCCs and suggests that specific variations in the embryological development of such tracts may result in less predictable relationships with surrounding structures (4). One theory postulates an "excess proliferation and invagination" of one pouch resulting in fusion of surrounding structures thought to be exclusively of a different subtype (4).

Conclusions

- BCCs can exhibit hybrid features resulting in a less predictable anatomic course.
- Careful review of imaging to determine the path of the tract and proximity of key structures is crucial.
- Facial nerve monitoring during excision of any BCC in close proximity to the parotid gland is recommended.

References