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CASE REPORT

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First branchial cleft sinus with duplicate external auditory canal: A case report

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Abstract

First branchial cleft cysts are rare congenital abnormalities of the lateral neck. These anomalies can be a source of recurrent infection and require surgical excision as definitive treatment. This case report details the diagnosis and treatment of a first branchial cleft sinus with an associated duplicate external auditory canal.

KEYWORDS

auditory canal abnormalities, branchial cleft, congenital anomalies, craniofacial abnormalities, otorhinolaryngologic surgical procedures

1 | INTRODUCTION

Congenital first branchial cleft anomalies are rare diagnoses that present as cysts, sinuses, or fistulae in the lateral neck between the sternocleidomastoid muscle and mandibular angle, below the external auditory canal (EAC), and/or above the hyoid bone.[1] The developmental defects are due to persistence of the branchial apparatus, a transient embryologic structure consisting of six mesodermal arches, five ectodermal clefts, and five endodermal pouches. This structure forms between Weeks 4 and 7 of fetal development until the arches fuse. The failure of this membrane to fuse results in the formation of persistent tracts or cysts depending on the degree of closure. Given that this fusion proceeds from the hyoid region toward the ear, it is more common for defects to occur in the periauricular region. These anomalies arise prior to the upward migration of the facial nerve and formation of the parotid gland, resulting in variable anatomical relationships between nerve and branchial remnant.[2] With an estimated incidence of one per million individuals per year globally, first branchial cleft anomalies comprise <8% of all branchial anomalies.[3] The condition can be asymptomatic for many years and is often diagnosed after becoming infected. They commonly present with swelling and drainage from a sinus or fistula tract in the lateral neck or EAC. Surgical resection is the standard of care to prevent recurrent infections and correct cosmesis. This report details the excision of a first branchial cleft cyst and duplicate EAC in a patient who experienced recurrent infections.

2 | CASE REPORT

A 21-year-old woman presented via telemedicine visit with a history of a left first branchial cleft remnant diagnosed at age 5. Over the past year, she experienced recurrent infections with swelling and drainage from her left cheek skin near the angle of the mandible. This was treated with incision, drainage, and antibiotics for patient comfort. However,

she was informed that surgical excision was the only definitive treatment to prevent future infections. A computed tomography (CT) scan was performed to evaluate for other possible parotid pathologies including neoplastic processes. Imaging revealed a 1.0 × 0.4 × 1.1 cm hypodense lesion along the surface of the parotid gland anterior and inferior to the EAC consistent with a first branchial cleft remnant. The patient underwent surgical excision as described below:

Electrodes were placed to monitor the left orbicularis oris and oculi muscles. After identifying a sinus/fistulous tract to the preauricular skin anterior and inferior to the left tragus, a preauricular incision was made and extended postauricular and into the occipital hairline. A skin flap was elevated in the face, preserving the posterior branch of great auricular nerve in the plane of the superficial musculoaponeurotic system (SMAS). The skin surrounding the draining tract was excised in an ellipse (Figure 1), and a lacrimal probe was placed in the draining tract (Figure 2). A pinhole sinus was cannulated superficial to the parotid fascia, leading to a cartilaginous duplicated EAC running parallel to the normal canal. The canals were separated by a party wall of hair-bearing skin without cartilage (Figure 3).

The facial nerve main trunk was identified in order to safely follow the duplicated ear canal medially. The parotid fascia was elevated off the normal EAC and sternocleidomastoid muscle. The posterior digastric belly and tympano-mastoid suture line were identified. Additional parotid fascia was divided to reveal the main trunk of the facial nerve (Figure 4).

The duplicated EAC was followed medially to a blind end lateral to the facial nerve main trunk, confirming this to be a first branchial cleft sinus rather than fistula. After dissecting down to the blind end, the party wall skin was excised such that the skin of the duplicated canal was incorporated into the normal EAC, forming a common canal (Figure 5). The facial nerve stimulated robustly at 0.5 milli-Amps. The normal ear canal was examined thoroughly using a binocular microscope, confirming no evidence of a fistula. The entire duplicated tract was removed, and the incisions were closed.

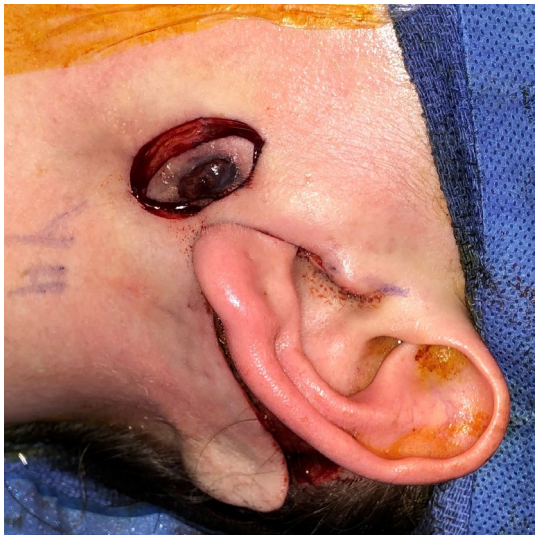

FIGURE 1 External sinus fistula with surrounding excised skin. A preauricular incision is extended into the postauricular region and into the occipital hairline

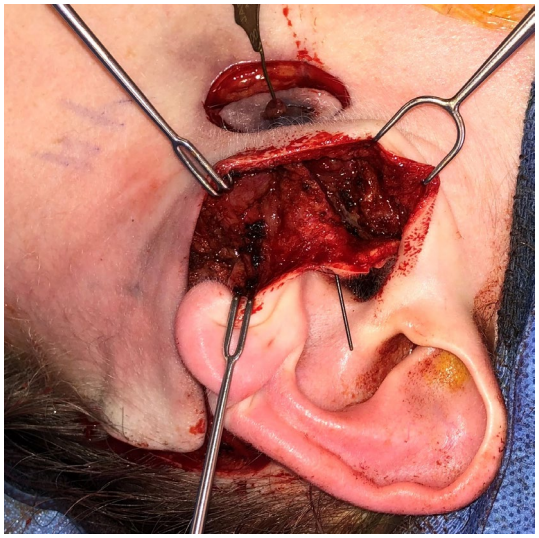

FIGURE 2 Lacrimal probe cannulates the draining sinus into the duplicated external auditory canal

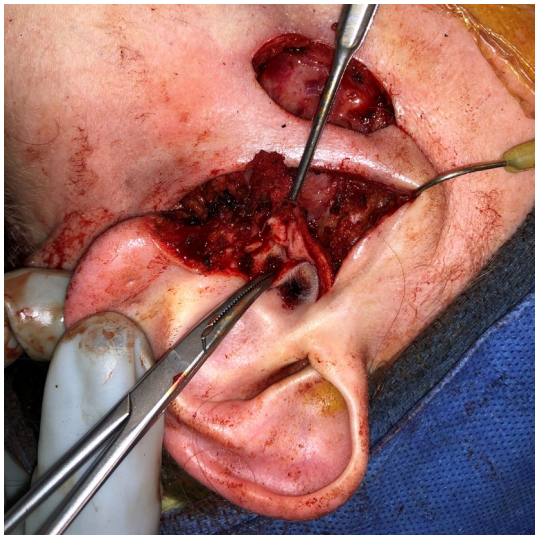

FIGURE 3 Duplicated external auditory canal (indicated by the mosquito hemostat) runs nearly parallel to the normal external auditory canal

3 | DISCUSSION

First branchial cleft anomalies can present from early childhood to late adulthood, with an average age of

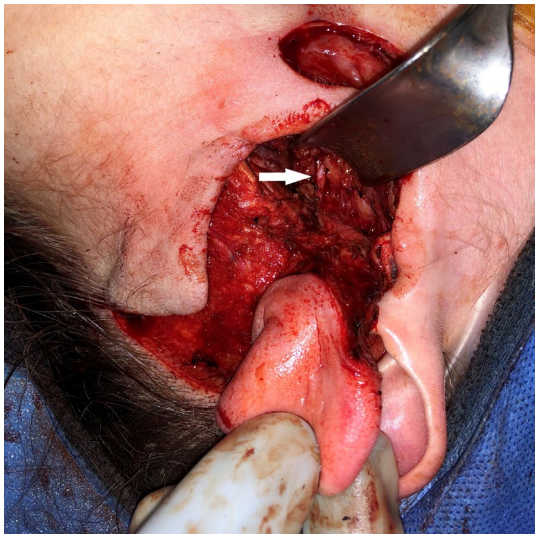

FIGURE 4 Surgical region after the dissection of the branchial cleft sinus including duplicated external auditory canal. The main trunk of the facial nerve is visualized (white arrow)

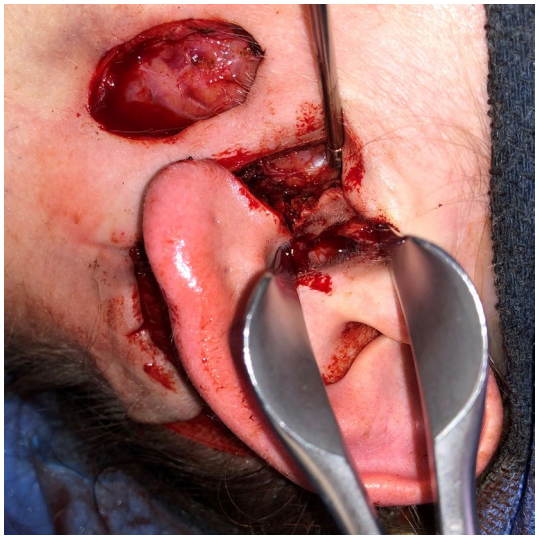

FIGURE 5 Both the normal and duplicated external auditory canals are visualized following the dissection of the common septum. The duplicated canal now shares a common lumen with the normal external auditory canal

diagnosis at 19 years. Definitive treatment is often delayed several years following symptoms due to misdiagnosis with limited treatment.[3] The type of remnant—cyst, sinus, or fistula—may also influence the age of diagnosis, with draining tracts presenting more commonly in early childhood.[2]

First branchial cleft anomalies are subdivided into two types based on histology and clinical presentation as described by Work in 1972.[4] Type 1 anomalies are cystic masses lined by squamous epithelium, commonly located near the ear and extending to the postauricular crease. Type 2 anomalies contain adnexal skin structures or cartilage and may extend to the face, upper neck, or into the EAC. They may present as cysts, sinuses, or fistulae and can lie medial to the facial nerve. Both types may include a duplicated EAC.[5] These anomalies are often asymptomatic until they become infected, presenting with episodic swelling in the periauricular or upper lateral neck region. Patients may also experience drainage from the periauricular skin and/or EAC. Otitis externa may develop, leading to conductive hearing changes.[2]

Neck CT is often sufficient for visualization of these fluid-filled cysts and/or tracts, and to evaluate for other pathology. Higher-cost magnetic resonance imaging (MRI) can also be used with excellent soft-tissue resolution.[3] Imaging has limited ability to define the anatomical relationship between the remnant tract and the facial nerve. This relationship varies between cases, with some reports noting fistulous tracts between or medial to facial nerve branches.[2] While not all branchial cleft anomalies are located near facial nerve branches, this should be considered during preoperative planning, and surgeons should prepare to identify and protect these branches intraoperatively as indicated. Surgeons should have experience with facial nerve dissection and parotid surgery, and counsel patients appropriately on the complications of iatrogenic facial nerve injury.

4 | CONCLUSION

First branchial cleft remnants are rare findings that can lead to recurrent infections without adequate treatment. Complete surgical resection of these anomalies often results in eradication of symptoms with low recurrence rates. Patients with swelling or infections in the periauricular and upper lateral neck region should be evaluated by physical examination and anatomical imaging. Surgical excision is the standard of care, as medical treatment alone is unlikely to provide long-term resolution. Given that preoperative imaging may be limited in its ability to define the anatomical relationship between the branchial remnant and facial nerve branches, surgeons should be prepared to identify and protect the facial nerve intraoperatively, and counsel patients on facial nerve complications preoperatively.

ACKNOWLEDGEMENTS

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CONFLICT OF INTEREST

There are no conflicts of interest or funding to disclose.

AUTHOR CONTRIBUTIONS

Matthew Jorizzo served as a primary author of the case report. Ryan Li, MD served as an attending surgeon for the procedure, edited the main body of the case report.

CONSENT

Patient consent for the use of procedure information and photographs in this case report have been signed and collected in accordance with the patient consent policy of *Clinical Case Reports*. This report was reviewed by the OHSU IRB office, which deemed the activity as not research involving human subjects (IRB ID: STUDY00023202).

DATA AVAILABILITY STATEMENT

Data sharing is not applicable to this article as no new data were created or analyzed in this study. The data that support the findings of this study are openly available in Pubmed at: https://pubmed.ncbi.nlm.nih.gov/. The DOI for each article is listed in the references below.

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REFERENCES

1. Quintanilla-Dieck L, Virgin F, Wootten C, Goudy S, Penn E Jr. Surgical approaches to first branchial cleft anomaly excision: a case series. *Case Rep Otolaryngol*. 2016;2016:3902974. doi:10.1155/2016/3902974
2. Maithani T, Pandey A, Dey D, Bhardwaj A, Singh VP. First branchial cleft anomaly: clinical insight into its relevance in otolaryngology with pediatric considerations. *Indian J Otolaryngol Head Neck Surg*. 2014;66(Suppl 1):271-276. doi:10.1007/s12070-012-0482-0
3. Chavan S, Deshmukh R, Karande P, Ingale Y. Branchial cleft cyst: a case report and review of literature. *J Oral Maxillofac Pathol*. 2014;18(1):150. doi:10.4103/0973-029X.131950
4. Work WP. Newer concepts of first branchial cleft defects. 1972. *Laryngoscope*. 2015;125(3):520-532. doi:10.1002/lary.25202
5. Goudakos JK, Blioskas S, Psillas G, Vital V, Markou K. Duplication of the external auditory canal: two cases and a review of the literature. *Case Rep Otolaryngol*. 2012;2012:924571. doi:10.1155/2012/924571

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