—Case Reports—

Large First Branchial Cyst Extending into the Parapharyngeal Space: A Case Report

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Cystic diseases of the salivary gland include mucous cysts and plunging ranula; cysts in the parotid region are rare. In this report, we describe a case of a first branchial cyst in the parotid region. The cyst extended into the parapharyngeal space and was repeatedly infected. The patient was a 35-year-old woman who presented to our hospital with a mass on the left lower ear. Imaging findings revealed a cystic lesion in the parotid region that extended to the parapharyngeal space. A yellowish slurry was aspirated on a percutaneous fine-needle aspiration biopsy. Cytology revealed a class II tumor. The patient initially showed signs of infection and was treated with intravenous antimicrobial agents. After the infectious inflammation had resolved, surgery was performed to resect the cyst. The infection did not recur postoperatively. A large first branchial cyst extending into the parapharyngeal space, which communicates with Stensen's duct, is rare. Care must be taken during surgery because of the complicated positional relationship between the first branchial cyst, parotid gland, and facial nerve.
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Key words: parotid gland, cystic lesion, infection, Stensen's duct, branchial cyst

Introduction

Cystic diseases of the salivary gland include mucous cysts and plunging ranula; cysts in the parotid region are rare. In this report, we describe a case of parotid cyst with a large extension into the parapharyngeal space and recurrent infection, and discuss the relevant literature.

Case Report

A 35-year-old woman presented with swelling of her right ear. Physical examination revealed hard, elastic swelling affecting the lower right ear. Her medical and family histories were unremarkable. She first noticed the swelling in September 2006 and visited the otorhinolaryngology department of a local hospital. In November 2009, she was diagnosed as having an enlarged mass near the lower right ear, which caused pain and difficulty with mouth opening. On the same day she was referred to our department for examination and treatment.

The initial examination revealed redness, swelling, and tenderness in the right hypopharynx and difficulty with mouth opening (**Fig. 1**). No swelling of the pharynx or cranial nerves was observed. The cervical lymph nodes were not palpable, and no obvious external auditory canal fistula was observed.

Contrast-enhanced computed tomography (CT) showed a 35-mm lobulated mass with contrast enhancement around the shallow and deep lobes of the parotid gland. Contrast-enhanced magnetic resonance imaging revealed a lesion that was hypointense on T1-weighted imaging and hyperintense on a T2-weighted imaging, with enhancement of the surrounding tissue, suggesting that inflammation had spread toward the parapharyngeal space and skull base (**Fig. 2**).

Blood testing (initial visit) showed a normal serum

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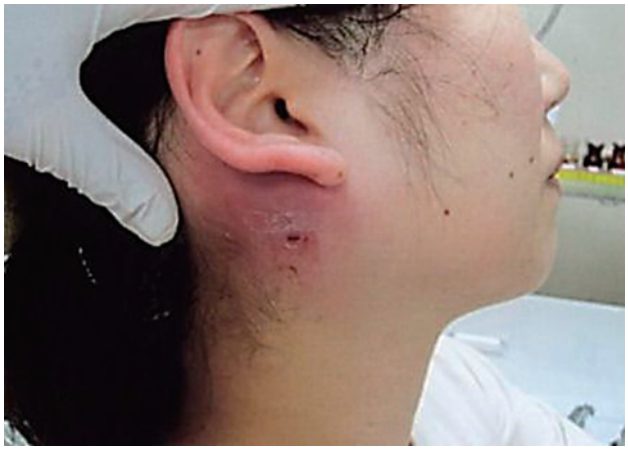

Fig. 1 Initial findings
The patient had erythema and swelling below the right ear.

amylase level; however, white blood cell concentration was 11,200/μL (neutrophils, 73.9%) and C-reactive protein level was 0.5 mg/dL, indicating a mild inflammatory reaction.

Atheromatous contents were aspirated by puncture aspiration cytology, and squamous cells were observed against a background of bacteria and neutrophils. No malignant findings, such as atypical cells, were observed, and the patient's tumor was classified as class II.

After admission, she was treated with intravenous piperacillin and clindamycin. The patient was considered to have an epithelial cyst infection, and surgery was planned after the inflammation had subsided. Infection continued to recur until surgery and she developed acute suppurative parotitis requiring hospitalization. She was treated with intravenous antibiotics to control the infec-

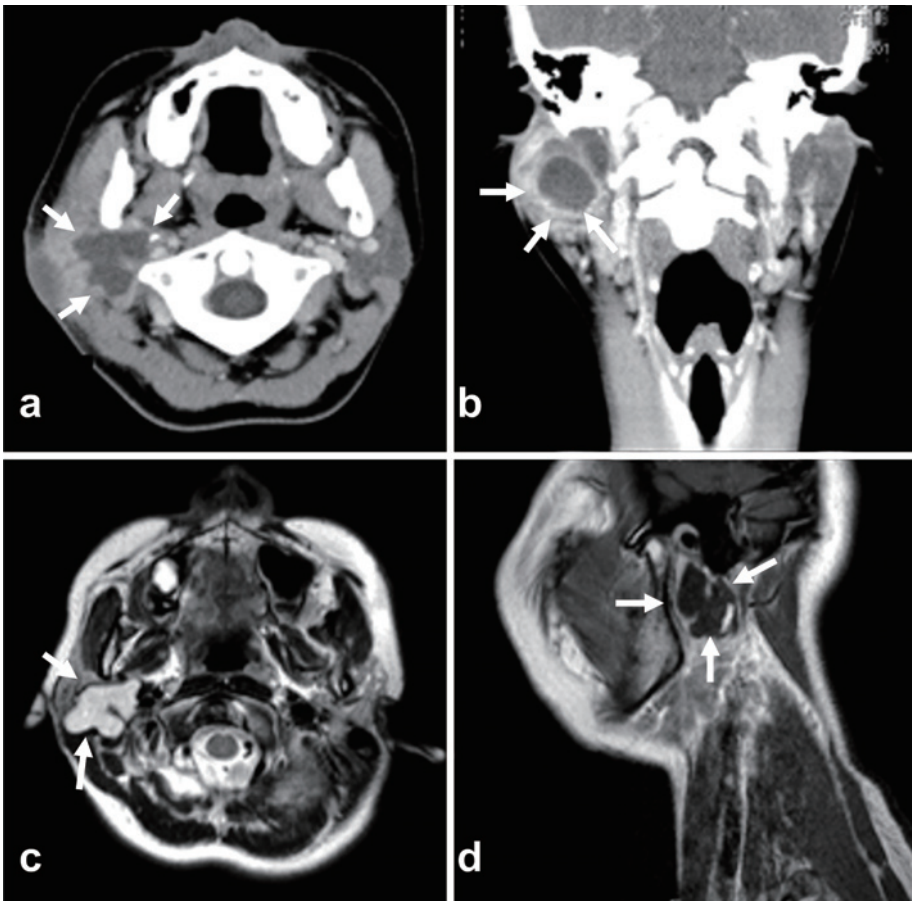

Fig. 2 Initial imaging findings
a: Contrast CT (horizontal section). b: Contrast CT (coronal section). c: MRI T2-weighted image (horizontal section). d: MRI Gd contrast T1-weighted image (sagittal section). a and b: A 35-mm lobulated mass with a contrast effect was observed around the parotid gland from the superficial to the deep lobes (white arrows). c and d: A lesion that is hypointense on a T1-weighted image and hyperintense on a T2-weighted image is in contact with the external auditory canal cartilage. Inflammation extends into the surrounding area (white arrows).
CT, computed tomography; MRI, magnetic resonance imaging.

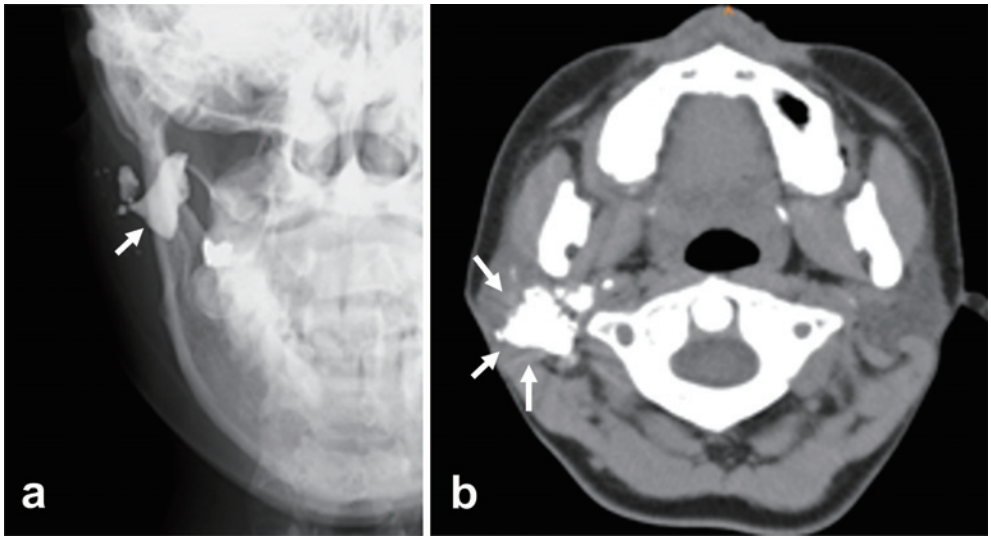

Fig. 3 Images from the CT test
a: Sialography of the right parotid gland. b: Simple CT (horizontal section) after sialography of the right parotid gland. a and b: retention of injected contrast medium in the cyst (arrows).

tion. During hospitalization, pus discharge was observed from the orifice of Stensen's duct into the oral cavity, and salivary gland duct angiography was performed to investigate the possibility of a cystic lesion communicating between the Stensen's duct and the mass. Subsequent CT revealed retention of the contrast agent in the cyst, which suggested communication between Stensen's duct and the cyst (**Fig. 3**).

The patient underwent surgery in late October 2010. A transparotid approach—with additional lateral mandibular dissection, if necessary—was adopted for the surgery. A Y-shaped incision was made at the side of the head below the ear, and the procedure was performed as in parotid tumor resection. The main trunk of the facial nerve was identified, and all branches were exposed. The mass extended from the deep lobe of the parotid gland toward the parapharyngeal space, and the facial nerve and mass crossed each other (**Fig. 4**). Mild adhesions were observed between the trunk of the facial nerve and its branches, and between the mass and normal parotid tissue, but dissection of the mass was possible. To obtain a clear view of the deep surgical field, the posterior fossa of the mandible was widely expanded with a retractor; osteotomy of the mandible was unnecessary. There was no obvious communication between Stensen's duct and the mass, and the mass was completely removed with good preservation of the facial nerve.

Histopathological findings revealed a cystic structure covered by stratified squamous epithelium with a granular layer. The cysts showed fibrosis and infiltration of lymphocytes, plasma cells, and neutrophils into the epithelium. No lymphoid follicles were observed (**Fig. 5**).

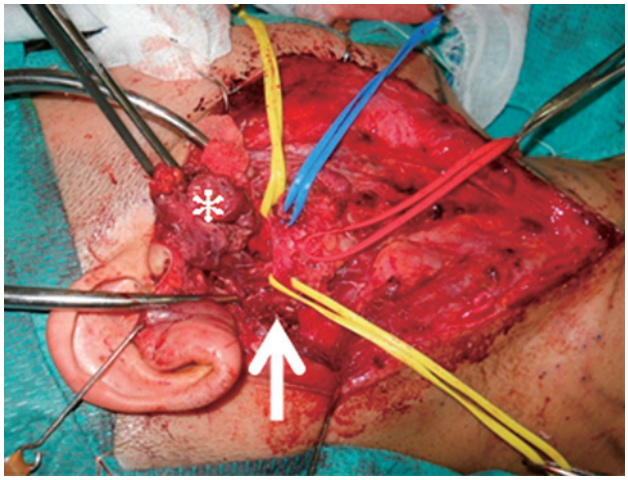

Fig. 4 Intraoperative findings
The mass crossed the facial nerve and was located in the deep lobe of the parotid gland along the external auditory canal. Arrow: main trunk of the facial nerve; * mass.

Facial nerve palsy, mainly in the upper main branch, was observed immediately after surgery; however, the patient recovered completely in approximately 2 months, and no recurrence has been observed to date.

This case report complies with the principles of the Declaration of Helsinki. Written informed consent was obtained from the patient.

Discussion

Cystic lesions that occur near the parotid gland and are

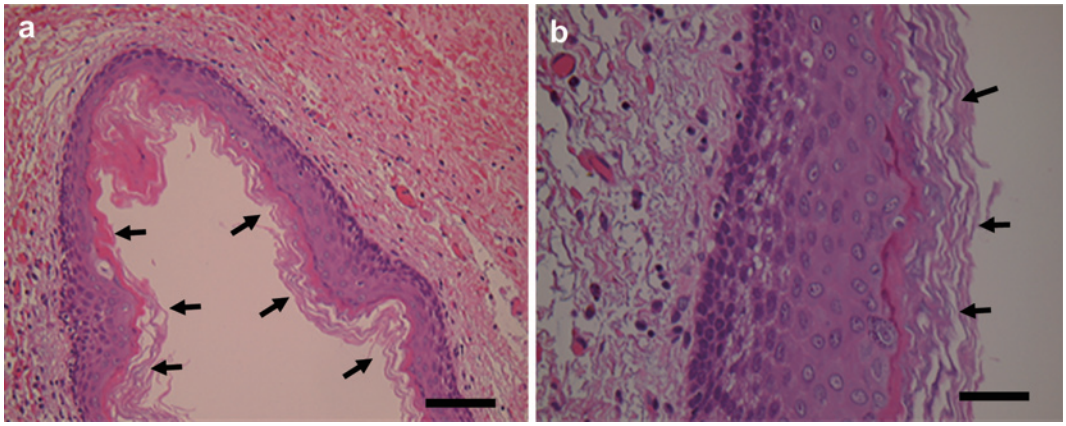

Fig. 5 Histopathological findings
a and b: Cystic structures coated with stratified squamous epithelium (black arrows). a: ×100, b: ×400. Scale bar = 500 μm.

coated with stratified squamous cells are classified as branchial, lymphoepithelial, and epidermoid cysts[1].

Branchial cysts are caused by the remnants of branchial clefts from the embryonic period or because of the inadequate closure of such clefts. A first branchial cyst is a rare condition that occurs at the boundary between the first and second branchial clefts. It is typically a pus-discharging cyst or fistula that extends from the lower mandible, along the inferior margin of the mandible, to the external auditory canal and auricle. Although present at birth, these cysts are often inconspicuous until puberty. The normal epidermis matures, and secretions from the epithelium lining the cyst increase in response to puberty, which may cause the cyst to enlarge and become infected. A first branchial fistula is easy to diagnose when a fistula is present. However, when it is a cyst, it must be differentially diagnosed from a parotid tumor on the basis of the site of origin. Symptoms of first branchial cysts include recurrent parotid abscesses and otorrhea. Hildebrant first reported branchial cysts in the parotid region in 1895, and approximately 100 cases have been reported. Branchial cysts in periparotid masses are very rare (frequency, 0.6%-2.9%)[2,3]. The locational relationship between the first branchial cleft and facial nerve is not constant, because the facial muscles and nerve arising from the second branchial arch move to cover the face in the first branchial arch region. In some cases, cysts pass through the shallow layer of the facial nerve and reach the deep layer, depending on the timing and site of the development of the first-gill cleft[4].

Lymphoepithelial cysts are solitary, unilateral masses that mainly develop in the parotid glands. Most affected patients are asymptomatic, except those with neck swelling due to a painless mass. The cyst wall is composed of squamous cells, columnar epithelium, and goblet cells with dense lymphoid tissue. Although considered a branchial arch-derived cyst, it is more likely to be caused by cystic degeneration of the epithelium of the salivary gland, which strays into the parotid gland or surrounding lymph nodes[5]. Even now, the differences between these two mechanisms are not clear. Bhaskar and Bernier[6]. diagnosed 452 of 468 branchial arch-derived cysts in the head and neck region with lymphoid tissue around the cyst wall as lymphoepithelial cysts.

A dermoid cyst is formed when skin and epidermal tissues stray into the dermis or subcutis because of congenital histological abnormalities or trauma. Histologically, the inner wall of the cyst is covered with a multilayered squamous epithelium, and the cyst usually has skin appendages. Such cysts are called dermoid cysts, and those without skin appendages are called epidermoid cysts. The pathological features of these diseases are identical, which makes a definitive clinical diagnosis difficult.

The present cyst was diagnosed as a first branchial cyst because of the recurrent parotid abscess extending along the external auditory canal to the deep lobe of the parotid gland and parapharyngeal space with slight adhesions; these findings are consistent with the pathological features of a first branchial cyst. During surgery, the cyst was located medial to the facial nerve while intertwining with it; however, preservation of the facial nerve was possible. In this case, communication between the cyst and Stensen's duct may have been caused by repeated inflammation that diffused into the surrounding tissues and organs, resulting in the weakening of the cyst

wall. Histopathologically, lymphocytes and neutrophils were observed around the stratified squamous epithelium within the cyst wall. This is an extremely rare case, as there have been no reports of communication between a parotid cystic lesion and Stensen's duct.

In summary, we described a case of a parotid cyst extending from the deep lobe of the parotid gland to the parapharyngeal interspace with recurrent infection. The cyst probably originated from a first-gill cleft cyst and was connected to Stensen's duct because of recurrent inflammation during the course of the disease, making this a very rare case. The positional relationship between a first branchial cyst, parotid gland, and facial nerve can be complex and requires careful attention during surgery.

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