CASE REPORT

Facial extension of ear pathology: infected cholesteatoma causing a parotid abscess

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SUMMARY

A man in his early 30s presented with right-sided preauricular swelling and facial oedema. He had a history of acid injury to his right ear as a child resulting in pinna deformity and subsequent blind sac closure of the external auditory canal. Imaging showed abnormal ear anatomy and abnormal density of the right parotid gland. Antibiotic therapy prevented progression but did not resolve the symptoms. Therefore, the infected area was surgically drained. This showed an underlying cholesteatoma, a benign but locally destructive condition where keratinising squamous epithelium grows in the middle ear and mastoid. The infected region was drained and the cholesteatoma was excised. This led to full resolution of the infection. The patient is awaiting a follow-up diffusion-weighted MRI. This case was unusual as the disease had extended beyond the ear and we therefore wish to alert clinicians to cholesteatoma as a possible cause of facial swelling.

BACKGROUND

Adult patients commonly present to primary care physicians, dentists and emergency departments due to facial swelling that is caused by infection. Though this is most commonly due to salivary gland, dental, paranasal sinus or external ear pathology, the close proximity of the middle ear and mastoid should prompt clinicians to investigate these sites as possible sources of infection. This is especially relevant in the context of abnormal ear anatomy as was seen in this case.

CASE PRESENTATION

A male patient in his early 30s presented to our department due to right facial swelling and pain. His symptoms started 2 days previously when he noticed the pain immediately anterior to the right ear. This became associated with swelling and dizziness over the course of the first day. The patient saw his general practitioner the following day. Other than dizziness and preauricular swelling, no new otological symptoms were noted and no otological signs were elicited at this review. Diclofenac and flucloxacillin were started for a presumed infection.

As these medications did not improve his symptoms, he visited an out-of-hours primary care clinic later that day and received additional analgesia (co-dydramol) as well as a change of antibiotics to co-amoxiclav. These medications improved the facial pain, though the dizziness deteriorated on the subsequent morning to become associated with vomiting, inability to eat or drink and inability to

walk. These symptoms prompted admission on presentation to our service.

The patient's medical history included ear surgery in his native country following an acid attack at the age of 6 months. This resulted in deformity of the right external ear as well as closure of the right external auditory canal and loss of right-sided hearing. Owing to the stability of his condition and lack of symptoms other than hearing loss, the patient had not sought ear, nose and throat referral on immigration to the UK. On examination, the right parotid region was found to be exquisitely tender, swollen, erythematous and warm to touch. The only associated sign was nystagmus to the left. There was no other abnormality of note or evidence of intracranial complication.

INVESTIGATIONS

Initial blood tests revealed a white cell count of $11.8 \times 10^9/L$, a raised neutrophil count ($10.2 \times 10^9/L$) and a C reactive protein level of 66 mg/L. An initial ultrasound scan showed enlargement of the right parotid gland. As the patient's symptoms did not improve after 48 h of intravenous antibiotic therapy, a CT scan of the head was performed. This showed a rim-enhancing fluid collection that extended from the superior aspect of the right parotid gland into the expanded right mastoid through a breach in the wall of the inferior mastoid bone. The entire right middle ear and mastoid was opacified with abnormal soft tissue. The ossicular chain was intact but there was associated bony erosion of the facial nerve canal and lateral semicircular canal (figure 1).

DIFFERENTIAL DIAGNOSIS

There is a wide range of infective, inflammatory and neoplastic causes for parotid swelling. Benign epithelial neoplasms of pleomorphic adenoma and Warthin's tumour predominate¹ and these cases tend to present indolently. More acutely, as in this case, bacterial or viral infection of the parotid gland parenchyma as well as infection of the intraparotid and periparotid lymph nodes can give rise to parotid abscess formation.² The most common causative bacterium is *Staphylococcus aureus* followed by anaerobes including Gram-negative bacilli, *Fusobacterium* spp, *Peptostreptococcus* spp and *Streptococcus* spp (including *S. pneumoniae*).³ Despite widespread measles, mumps and rubella vaccination, mumps still remains an important viral cause of parotitis due to an 86.3% vaccine effectiveness^{4 5} though this remains, predominantly, a problem of childhood.

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Figure 1 CT scan of the head from initial admission. (A) Axial postcontrast CT scan showing a right-sided 2.2 cm×1.2 cm by 1.8 cm rim-enhancing fluid collection. (B) Coronal reconstruction of the same CT head scan showing extension of the fluid collection into the mastoidectomy cavity. (C) Bone-windowed axial CT image showing the opacified thin-walled mastoid cavity created by cholesteatoma. Ossicles are noted in the opacified right middle ear cavity (closed black arrowhead). (D) Coronal bone-windowed CT image of the mastoid showing soft tissue eroding the medial wall and causing lateral semicircular canal dehiscence (open right arrowhead). There is also dehiscence of the mastoid segment of the facial nerve (open black arrowhead). Closed white arrowhead, rim-enhancing fluid collection in parotid region and asterisk, right mastoid cavity.

Bacterial parotid infection is more commonly found in elderly, dehydrated patients who are intubated or diabetic.⁶ This did not correlate with our patient who was young and otherwise well. Similarly, the majority of patients with cholesteatoma have a history of ear discharge, otalgia and hearing loss⁷ which our patient could not have recognised due to his previous ear problems and therefore provided no assistance to his diagnosis.

More relevantly, a significant minority of patients with cholesteatoma (13.2% according to one study) recall ear trauma or surgery. This fact, coupled to the CT scan results, raised the possibility of cholesteatoma underlying our patient's acute parotid problem. Though there are case reports of parotid cholesteatoma,⁸ we are not aware of any previous cases where this has presented acutely as a parotid abscess.

TREATMENT

Following secondary review of the existing imaging by an otologist in a tertiary centre, drainage of the parotid abscess via a postauricular approach was attempted in order to improve the patient's acute infection. After initial incision and anterior reflection of the external ear, cholesteatoma was noted to be filling the entire external auditory canal, mastoid cavity and middle ear (confirmed on histology; figure 2A). This may have been caused by implantation of skin cells to the mastoid cavity at the time of the patient's childhood surgery. Complete excision of the cholesteatoma revealed a large amount of pus that was suctioned (figure 2B) and found to be entering the external auditory canal via a fistula tract through the anterior external auditory canal (fissure of Santorini) to the parotid region with continuity to the mastoid bone via an inferior mastoid breach (figure 2C).

Following suction of the external auditory canal and pressure on the parotid region, it was possible to fully drain the preauricular collection without the need for a preauricular incision that would have put the facial nerve at greater risk. The mastoid appearance was consistent with previous mastoidectomy and the facial nerve was clearly visible as the bony covering that usually protects the nerve as it crosses the middle ear had been completely eroded (ie, the facial nerve was dehiscent). The right middle ear ossicles were preserved on the preoperative and postoperative CT scan (figure 1C), though they were not seen at the time of surgery as the cholesteatoma was fully excised by careful dissection around the dehiscent facial nerve. Finally, leakage of perilymph (a fluid that fills part of the inner ear) was seen and found to originate from a dehiscent lateral semicircular canal. This was the presumed cause of the patient's vestibular symptoms/signs and was repaired in order to improve these symptoms and protect any residual inner ear function.

The patient's infection rapidly settled following drainage and it was possible to discharge him home to complete a course of oral ciprofloxacin. Microbiology testing of the intraoperative sample demonstrated *Haemophilus influenzae* to be the causative organism.

OUTCOME AND FOLLOW-UP

Despite the underlying pathology, the abscess has not recurred since drainage and the patient remains well with no facial nerve compromise at 1 year follow-up. A keloid scar developed at the operative site and was treated by triamcinolone injection. CT of the petrous temporal bones was performed 4 months postoperatively to exclude any residual collection and did not identify any new abnormality. The lateral semicircular canal repair appeared

Figure 2 Intraoperative microscopic photographs of abscess drainage via a right postauricular approach. A postauricular incision has been made and the pinna has been retracted anteriorly. (A) On initial inspection, keratinised tissue consistent with cholesteatoma was seen to completely fill the external auditory canal. (B) Removal of the visible cholesteatoma revealed copious pus which was subsequently suctioned. (C) An anteriorly placed fistula tract was found to connect the external auditory canal to the parotid region. A, anterior; P, posterior; S, superior; I, inferior; M, mastoid region; dotted line, outline of external auditory canal and mastoid cavity opening; and asterisk, location of fistula tract between external auditory canal and parotid region.

intact (figure 3). As all macroscopic cholesteatoma was excised at the time of surgery, a 'second-look' operation is not currently scheduled. Instead, a diffusion-weighted MRI is planned for surveillance.

DISCUSSION

There is a true paucity of evidence regarding the extension of cholesteatoma to the parotid region. Two relevant case reports that were found were not in the English language.^{9 10} The remaining papers focus on epidermal inclusion cysts following ear surgery.^{11 12} These related cases refer to cysts which occur when epidermal elements from the dermis are introduced to the parotid area by incisions for ear surgery or trauma, though the incision may have been made to access a cholesteatoma. Furthermore, these cases present subacutely with the absence of infection.¹²

More relevantly, the extension of a cholesteatoma to the parotid region has been reported and this report gives the sensible recommendation of formal ear examination in patients who present with parotid swelling in the context of previous, ipsilateral, ear surgery.⁸ However, it was not possible to follow this advice in the present case as the patient did not have a patent external auditory canal. This presentation was also unusual in that infection was the prompt for presentation rather than swelling.

This posed a unique challenge given that the facial nerve runs through the substance of the parotid gland following its complicated course and exit from the temporal bone. If a preauricular incision had been made to approach the abscess, the facial nerve would have been at risk. This is similar to the challenge posed by complicated parotid tumours. However, in this context, the CT findings supported a source of the problem within the temporal bone.

This CT was consistent with cholesteatoma due to erosion of the facial nerve canal and lateral semicircular canal, even though the ossicular chain was intact. MRI with non-echoplanar diffusion-weighted images was considered for more preoperative diagnostic information but it was not possible to obtain this study out of normal working hours. MRI with non-echoplanar diffusion weighted imaging (DWI) and postcontrast MRI sequences could have helped to characterise the abnormal soft tissue. Both cholesteatoma and abscess are rim-enhancing and demonstrate restricted diffusion with lower apparent diffusion coefficient (ADC) values than inflammation or fluid.^{13–15} A small study has described lower ADC values for abscess than cholesteatoma.¹³ Non-echoplanar DWI can also define the extent of primary cholesteatoma and help in planning the surgical approach.¹⁶

Given the various risks, the postauricular approach was deemed safest and also allowed greater access to the underlying ear disease. Any subsequent operation (if necessary) will take into

Figure 3 CT scan of the temporal bone 4 months postoperatively. (A) Axial non-contrasted CT scan at the level of the right mastoid cavity. (B) Coronal reconstruction of the same CT temporal bones. *, right mastoid cavity; open arrowhead, reossified right lateral semicircular canal; and closed arrowhead, persistent right facial nerve dehiscence.

account the extent of the cholesteatoma, the known dehiscence of the facial nerve canal, the perilymph leak repair and the fistula from the external auditory canal to the parotid region.

Learning points

- ▶ Acute parotid swellings can be secondary to underlying tumours or tracts.
- ▶ The facial nerve is at risk from such infections.
- ▶ Formal ear assessment by the ear, nose and throat team is required where there is a history of ipsilateral ear surgery.
- ▶ Full clinical assessment may not be possible in rare cases, and in these cases, imaging by both CT and MRI can provide further insight.

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Competing interests None.

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