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Case report of cholesteatoma recurrence with Bezold's abscess presenting as a deep neck infection

M. Lionello[1], R. Manara[2], L. Lora[3], I. Mylonakis[1], E. Fasanaro[1], F.B. La Torre[1], G. Ottaviano[1], A. Staffieri[1] and R. Marchese Ragona[1]

[1]Department of Neuroscience, Otolaryngology Section, University of Padua, Padua, Italy; [2]Neuroradiology, University of Salerno, Salerno, Italy; [3]Department of Pharmaceutical Sciences, University of Padua, Padua, Italy

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Abstract. *Case report of cholesteatoma recurrence with Bezold's abscess presenting as a deep neck infection*. Cervical masses are a common clinical finding, but differential diagnosis is often challenging. Acute neck swellings are often due to deep cervical space infections that have originated at oral or oropharyngeal sites. Deep neck infections originating elsewhere are not rare; however, they are difficult to diagnose, and their origins remain obscure in 20% of cases. Neck swellings that originate in the middle ear are very rare, with only a few reported in the scientific literature. Here we report an atypical case of Bezold's abscess caused by the recurrence of a middle ear cholesteatoma. In patients with neck swelling and a history of primary cholesteatoma of the middle ear, otolaryngologists should consider regional recurrence of disease a possibility even several years after the primary surgery.

Introduction

Cervical masses are common clinical findings that pose a challenge in terms of their differential diagnosis. Acute neck swellings are frequently due to deep cervical space infections, which are fairly common complications of oral or oropharyngeal inflammation. Such infections are characterized by high morbidity rates if not diagnosed promptly and can even lead to life-threatening complications. The incidence of deep cervical abscesses has declined with the widespread use of antibiotics in clinical practice. The most common causes of deep neck infections are inflammatory processes that originate at oral or oropharyngeal sites. Dental root abscesses in particular are a common cause of deep neck infections (39.5%), but the origin remains unknown in 20% of cases.[1] In such cases, comprehensive testing should be used to determine whether other less common causes may be responsible.

Case report

A 35-year-old Moldavian man was admitted to our otolaryngological department complaining of a swelling in the left upper laterocervical region. His skin showed local hyperemia and he had no fever. ENT examination showed a normal oral cavity and slight lateral swelling of the right pharyngeal wall. Micro-otoscopy disclosed normal sequelae of left canal wall-down tympanoplasty with open mastoidectomy, which the patient had undergone 20 years earlier in his home country. The patient had no history of smoking or drinking alcohol. He reported no infectious or rheumatological diseases and denied any exposure to tuberculosis. Blood tests revealed no leukocytosis, and C-reactive protein (CRP) levels slightly above normal (17.00 mg/L; n.v. 0-6 mg/L). Hemocultures and the QuantiFERON test were negative. Contrast-enhanced CT of the neck revealed a solid mass 31 × 20 mm in size in the left cervical region that was under the mandibular angle and close to the internal jugular vein (Figure 1a). The patient started intravenous (iv) antibiotic therapy with 3 g of sulbactam sodium + ampicillin sodium 3 times a day plus 500 mg of metronidazole 3 times a day, and corticosteroid therapy with 4 mg of disodium phosphate betamethasone once a day. This is in line with accepted protocols for the treatment of deep neck infections.[1] Since the neck swelling persisted with no clinical improvement after 12 days of antibiotic therapy, ultrasound-guided fine needle aspiration of the neck abscess was performed, and the cytology revealed non-specific lymph reticular hyperplasia. The patient's clinical condition slowly improved,

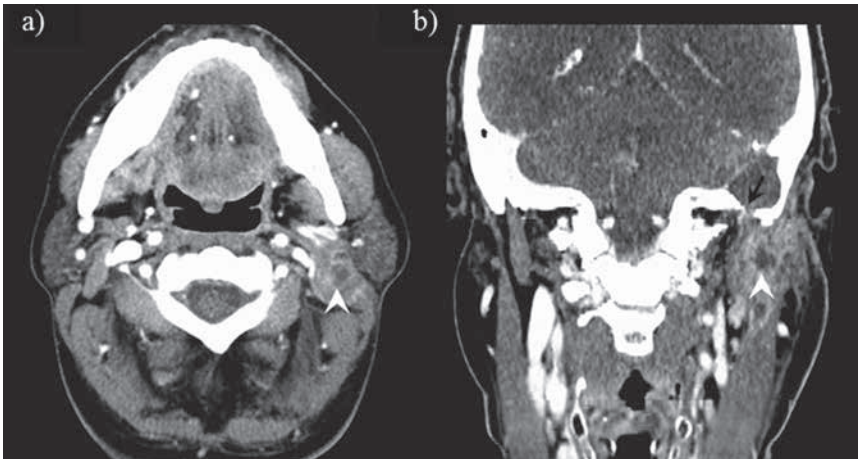

Figure 1
Head and neck CT
a) A contrast-enhanced axial CT image of the neck at the C1-C2 level shows soft tissue edema on the left side involving the sternocleidomastoid muscle, which appears swollen and unevenly enhanced; focal areas of necrosis are indicated by arrowheads. b) Coronal multi-planar CT reconstruction shows the contiguity between the mastoid mass and the inflamed area of soft tissue in the neck.

but the cervical mass recurred as soon as antibiotic therapy was discontinued. The recurrence of symptoms prompted us to change the antibiotic regimen to 2 g of ceftriaxone twice a day and 500 mg of metronidazole 3 times a day. Orthopantomography showed an abscess at the apex of tooth 37, which was subsequently removed. The patient's clinical condition improved and he was discharged, but the cervical mass recurred 4 days later. A new contrast-enhanced CT scan of the neck showed an abscess 10×14 mm in size in the para-pharyngeal space under the sternocleidomastoid (SCM) muscle with thrombophlebitis of the left jugular vein (Figure 1b). The patient underwent neck exploration on the same day, which revealed an abscess close to the apex of the mastoid adjacent to the deep cervical extremity of the SCM muscle. The abscess was drained. Antibiotic iv therapy was continued without anticoagulant therapy according to Kuppalli *et al*.,[2] and purulent discharge from the suction drain continued over the next 10 days. This unusual course prompted us to suspect that the abscess originated in the ear. The patient was subjected to a temporal bone CT, which showed sequelae of left canal wall-down tympanoplasty, no abnormal tissue within the neocavity (Figure 2a) and normal inner ear and labyrinthine structures. However, bone erosion that was not due to the earlier mastoidectomy was observed (Figures 2b, 2c). Because cholesteatoma was suspected, echo-planar diffusion-weighted (EPI-DWI) and contrast-enhanced magnetic resonance imaging (MRI) of the temporal bones was performed. This revealed a 2.8-cm cholesteatoma with cortical bone erosion of the mastoid apex and a small neck abscess in the contiguous deep cervical space (Figure 3, Figure 4). The patient underwent a radical modified mastoidectomy and new cervical exploration. The retro-auricular incision was extended to the neck along the anterior margin of the SCM muscle, disclosing the link between the apex of the mastoid and the adjacent neck abscess (Figure 1b). The postoperative course was uneventful, and the patient was discharged 7 days later. He remains disease- and symptom-free 12 months after surgery.

Discussion

In 1881, Friedrich Bezold first described an abscess deep in the muscle planes of the neck with suppuration from the mastoid tip cells that was secondary to acute mastoiditis. At the beginning of the 20th century, about one of every two patients with otitis media developed coalescent mastoiditis. The incidence of complications of acute suppurative otitis media decreased significantly after the introduction of antibiotics in routine clinical practice and by 1959 the incidence of this kind of complication was only 0.4%.[3] The management of Bezold's abscess is both medical and surgical, as both early antibiotic

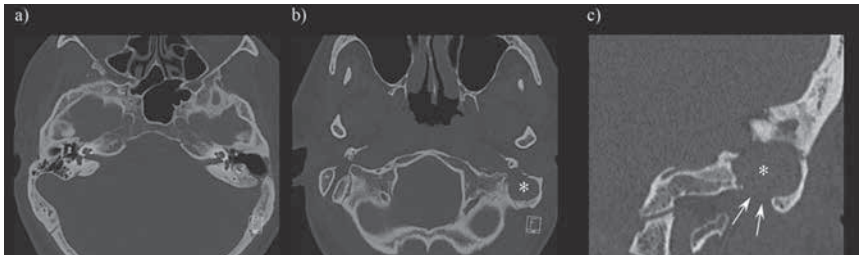

Figure 2
Petrous bone CT
a) An axial CT image of the internal acoustic canal shows sequelae of wall-down surgery in the left petrous bone. The cavity appears clean and well-aerated. b) An axial CT image of the lower part of the mastoid reveals a round, well-defined cavity in the left mastoid that is filled with isodense tissue (*). c) A coronal multi-planar CT reconstruction through the left mastoid bone shows a large bone defect (white arrows) in the inferior cavity wall.

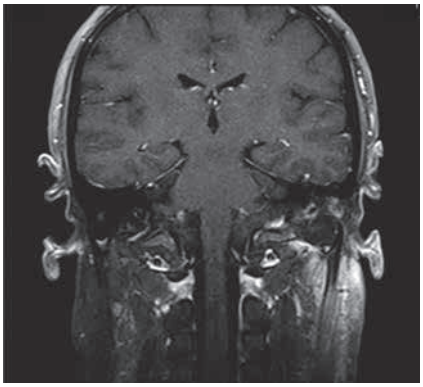

Figure 3
Head and neck MRI
A contrast-enhanced, fat-saturated T1-weighted coronal image shows enhancement of the left sternocleidomastoid muscle at its insertion in the mastoid bone.

therapy and timely drainage of the neck and the mastoid are needed for cure. Surgical exploration and drainage of a deep neck infection is mandatory for patients who fail to respond to parenteral antibiotics within the first 24 to 48 hours.[1] Familiarity with the clinical presentation of this uncommon condition may help prevent diagnostic delays and enable prompt therapy.[4]

The case presented here shows that Bezold's abscess can be caused by the recurrence of cholesteatoma even many years after primary surgery. Classic Bezold's abscess is described as a complication of acute mastoiditis; however, here there was a chronic process underlying its occurrence, making this clinical presentation very unusual.[5] Because of the tendency of cholesteatoma to invade nearby structures, cholesteatoma patients are at higher risk of complications than those with other types of otitis

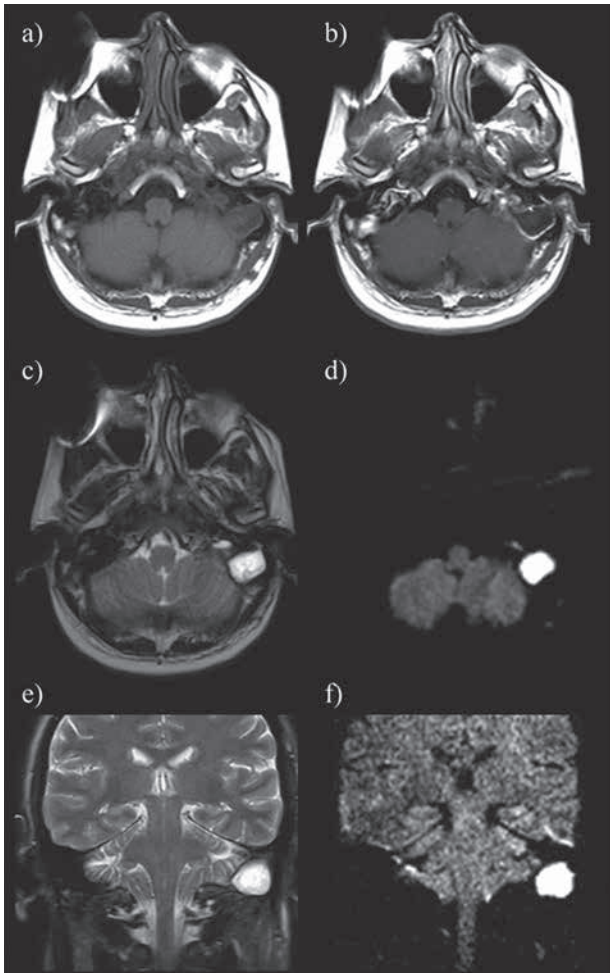

Figure 4
Head and neck MRI.
T1-weighted (a), contrast-enhanced T1-weighted (b), T2-weighted (c) and EPI-DWI (d) axial images of the mastoids show a large, T1-hypointense and T2-hyperintense left mastoid mass with subtle ring-like enhancement and marked hyperintensity on DWI, consistent with cholesteatoma. T2-weighted (e) and echoplanar DWI (f) coronal images of the intra-mastoid cholesteatoma.

media because of its tendency to invade structures nearby.[6] It is not unusual for a cholesteatoma to recur several years after primary surgery.[7] Otolaryngologists need to be aware that they may encounter unexpected clinical conditions when dealing with benign processes such as cholesteatoma. Especially in patients with a history of surgery of the middle ear, changes to the anatomical structures often have unexpected consequences. In our patient, prior mastoidectomy enabled the cholesteatoma to spread towards the mastoid apex. Although this is not the first reported case of cholesteatoma spreading to the neck,[8,9,10] our patient presented with a perfectly well-aerated tympanic cavity and cervical symptoms. On CT scan, the metaplastic mucosa could be considered erroneously to be non-pathological tissue. When in doubt, enhanced temporal bone T1-, T2- and diffusion-weighted MRI (Figure 3, Figure 4) is mandatory to rule out cholesteatoma of the middle ear. Although non-EPI DWI is considered a more sensitive way to detect cholesteatoma than EPI-DWI, especially when the lesion is less than 5 mm, nonetheless non-EPI DW sequences are time consuming compared to EPI-DWI, require newer MR scanners and do not add important information when dealing with lesions greater than 5 mm, such as this case.[11]

Conclusions

The differential diagnosis of a cervical mass in patients treated previously for primary cholesteatoma of the middle ear should include consideration of the possibility of a regional recurrence of cholesteatoma, even years after the primary surgery.

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Marco Lionello, M.D.
Department of Neuroscience, Otolaryngology Section
University of Padova
Via Giustiniani 2
35128 Padova, Italy
Fax: +39 049 8213113
E-mail: marcolionello@email.it