CSF otorrhea complicating temporal bone osteoradionecrosis in a patient with nasopharyngeal carcinoma

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Abstract
A 44-year-old Chinese man with a history of nasopharyngeal carcinoma that was treated with radiotherapy presented with fluid in the middle ear. We performed a myringotomy and subsequently made a diagnosis of cerebrospinal fluid (CSF) leakage secondary to osteoradionecrosis of the temporal bone. To the best of our knowledge, this is only the second reported case of an otogenic CSF leak resulting from osteoradionecrosis of the temporal bone. This case highlights the controversial role of myringotomy in the management of CSF otorrhea.

Introduction
Nasopharyngeal carcinoma is a common malignancy among the Chinese, and it is one of the ten most common cancers overall in Singapore. Patients typically present with a posterior-triangle cervical lymphadenopathy and ear blockage secondary to otitis media with effusion. Physical examination usually reveals the presence of a nasopharyngeal mass on fiberoptic endoscopy. Biopsy analysis confirms the diagnosis.

Because radiotherapy is the mainstay of treatment for nasopharyngeal carcinoma, the role of the otolaryngologist primarily involves its diagnosis, follow-up, treatment of both regional metastasis (via neck dissection) and local metastasis (via nasopharyngectomy), and treatment of middle ear effusions. However, because radiotherapy is associated with poor tissue healing, radiation-induced auditory neuropathy, and middle ear effusion, we are faced with deciding whether to perform a myringotomy, which will transform a poor-hearing dry ear into a poor-hearing discharging ear.

In this article, we describe a case of an otogenic cerebrospinal fluid (CSF) leak secondary to osteoradionecrosis of the temporal bone. To the best of our knowledge, this is only the second such case reported in the literature.

Case report
A 44-year-old Chinese man presented with a 3-week history of otalgia and right ear blockage. Four years earlier, he had been diagnosed with a poorly differentiated T1N2M0 nasopharyngeal carcinoma, and he underwent a course of concurrent chemotherapy and radiotherapy.

Physical examination detected mild tympanosclerosis bilaterally and evidence of right middle ear effusion. Findings on a neurologic examination were unremarkable, no cervical lymphadenopathy was evident, and the anterior and posterior nasal spaces were essentially normal. A right myringotomy yielded a clear, persistent, pulsatile discharge. A biochemical analysis revealed that the discharge was CSF.

Computed tomography (CT) of the posterior nasal space and temporal bone demonstrated opacification of the right tympanic cavity and a noticeable fluid level. CT also suggested the early stage of erosion of the superior margin of the petrous temporal bone anteriorly, just in front of the middle ear. The bony cortical margin was not well visualized. Otherwise, there was no evidence of a recurrence of his cancer in the postnasal space and no visible abnormality at the bony base of the skull. Finally, no cervical lymphadenopathy was observed.

In view of the persistent CSF otorrhea, an exploration of the middle ear cavity and closure of the perilabyrinthine CSF leak was performed. Intraoperative findings included a perilabyrinthine CSF leak from the air cells around the semicircular canals. Also, a tegmen defect was noted superiorly. We performed a right subtotal petrosectomy, a repair of the tegmen defect with a temporal fascia and fat graft secured with fibrin glue, and a blind sac closure of the right external auditory canal.

Postoperatively, the patient recovered uneventfully, and
he was discharged on postoperative day 3. He remained well at the 9-month follow-up evaluation.

Discussion
Most cases of nasopharyngeal carcinoma are treated with radiotherapy. One of the several complications of radiotherapy in these cases is osteoradionecrosis of the temporal bone, which receives a high dose of radiation over the course of therapy. Two mechanisms have been suggested to explain the occurrence of osteoradionecrosis in these patients:

- One suggestion is that oblitative vasculitis causes a direct radiation-induced avascular necrosis of the bone. This is more likely to occur in the presence of tumor involvement. There is a positive relationship between the size of the radiation dose and the degree of necrosis.
- Another explanation is that infection plays a role in aggravating osteoradionecrosis.

Ramsden et al classified osteoradionecrosis of the temporal bone as either localized or diffuse. In localized osteoradionecrosis, the disease is generally confined to the external auditory canal, and symptoms manifest according to the site and stage of the disease. Patients with localized disease usually present with local dermatitis, otalgia, and especially otorrhea.

In contrast, diffuse osteoradionecrosis extends beyond the temporal bone to the base of the skull and its surrounding structures. Affected patients present with more severe symptoms, including a profuse and pulsatile otorrhea and significant pain. The diffuse form is associated with a greater likelihood of complications, including trismus, intracranial infection, facial nerve palsy, labyrinthitis, chronic mastoiditis, CSF leak, and internal carotid artery aneurysm.

The case we have described features a rare but important complication of osteoradionecrosis of the temporal bone secondary to radiotherapy for nasopharyngeal carcinoma: otogenic CSF leak. To the best of our knowledge, only one other case has been previously reported. We suspected a CSF leak in our patient when the myringotomy yielded the clear, persistent, pulsatile discharge from the right ear. Biochemical analysis of the fluid confirmed the diagnosis. In patients with otogenic CSF leaks, glucose and protein levels are usually measured and isolation of \( \beta_2 \) transferrin is recommended.

Although most symptoms of osteoradionecrosis usually manifest many years following radiotherapy, symptoms secondary to its soft-tissue effects can present earlier. In our patient, the interval was 4 years.

In the radiologic evaluation of CSF otorrhea, noncontrast high-resolution CT is preferred over CT cisternography and radionuclide cisternography. The radiologic evaluation can be supplemented by multiplanar magnetic resonance imaging. However, because osteoradionecrosis usually distorts the anatomy, even high-resolution CT may not be sufficient; in such a case, surgical exploration is indicated.

CSF leakage can be treated surgically or nonsurgically. Nonsurgical treatment of temporal bone osteoradionecrosis entails regular aural toilet, topical antibiotics, and perhaps hyperbaric oxygen therapy. Other nonsurgical measures include bed rest, head elevation, and insertion of a subarachnoid drain. If nonsurgical treatment should fail, surgical intervention is indicated. Different surgical approaches have been described as definitive treatments, but a mastoid approach, a middle fossa approach, or a combination of both is recommended. In our patient, the decision to perform surgical closure was based on the intraoperative findings.

References