CASE REPORT

Lateral semicircular canal osteoma presenting as chronic postaural fistula

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SUMMARY

Temporal bone osteoma is an unusual pathology which can occur by birth or can be acquired and mostly involves the tympanomastoid segment of the temporal bone. Osteomas arising from the otic capsule are extremely rare, and there has been only one other report of a lateral semicircular canal osteoma in the literature. We report a similar case of an acquired lateral canal osteoma which presented as a chronic postaural fistula in an ear previously operated for paediatric cholesteatoma.

BACKGROUND

An osteoma is a benign mature growth of well-differentiated osseous tissue, which is a rare finding within the temporal bone.¹ Of osteomas found in the skull, temporal bone osteomas constitute only between 0.1% and 1%.^{1 2} Their causes include trauma, previous surgery, radiotherapy, chronic infection and hormonal factors with dysfunction in the hypophyseal gland.^{1 2} The most common site of temporal bone osteomas are the external auditory canal, followed by mastoid bone and, least commonly, the middle ear.³ There has only been one other case of an osteoma arising from the otic capsule/inner ear reported in the literature apart from this case report.⁴

CASE PRESENTATION

A 25-year-old woman presented to the otology department in a tertiary referral care centre with a 2-year history of a right postaural fistula discharging intermittently for 2 years. Eleven years earlier, she had undergone a combined approach tympanoplasty for a paediatric cholesteatoma involving the right middle ear cleft. Operative records from the original surgery described a cholesteatoma that was fully cleared from the attic/antral regions but that the lateral semicircular canal had been breached. It is unclear from the operation record whether this breach was secondary to disease or by surgical misadventure. Since the operation, there had been no hearing in the affected ear, and though she was initially dizzy following the procedure, she had made an excellent functional recovery, implying compensation following her 'dead' ear. Three years prior to presentation, the patient had suffered an episode of acute mastoiditis; however, a diffusion-weighted MRI scan indicated that there was no residual or recurrent cholesteatoma.

When seen in clinic, it was noted that the patient had recurrent mucosal discharge through

a tiny postaural sinus. There was some inflammation and erythema around the fistula site. This fistula tract in the recent past had shown granulations with mucous which grew mixed flora, partially responding to conservative management with antibiotics. Her neotympanum was intact but retracted with a clear attic. There was no history of discharge from the external auditory canal, and there was no posterior-superior canal wall sagging or cavity to connect to the postaural opening. Her presentation with an intact neotympanum with no evidence of middle ear infection was unusual for a person with postaural discharging fistula. She had no hearing or vestibular function in this ear since surgery and had coped well with her contralateral normal hearing ear. Her facial nerve function was normal and she had no vestibular signs.

INVESTIGATIONS

The current clinical history and scans suggested the most likely possibility of chronic otitis media. CT scan was reported as recurrent mastoiditis while the non-echo planar diffusion weighted MRI did not show convincing evidence of cholesteatoma. Further scrutiny of her CT scan by the otology team showed a solid bony mass deep to the fistula tract which seemed to extend up to the inner ear blocking the aditus region, with soft tissue opacification in the aditus while the rest of her middle ear seemed to be disease-free ([figure 1](#) and [figure 2](#)).

TREATMENT

The patient underwent mastoid exploration with excision of the fistula tract. During surgery, the tract was found to be superficial to the dense bone which had caused obstruction to the aditus and posterior tympanotomy with secondary chronic mastoiditis due to lack of ventilation, which led to the fistula formation. This tract was excised in an ellipse and the wound explored down into the mastoid. The remnant cortex was drilled off to enter the mastoid cavity to reveal a large solid otic capsule bony outgrowth arising from the lateral semicircular canal and extending towards the aditus and the previous posterior tympanotomy site. This was blocking access to the middle ear. The fistula was emerging posterior to this. The anatomy was difficult due to a dehiscence of the facial nerve near the second genu and the absence of surgical landmarks due to disease process and previous surgery.

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Figure 1 High resolution computed tomography scan with a black arrow pointing at the right lateral semicircular canal osteoma obstructing the view of the mastoid.

At the end of the procedure, the presumptive diagnosis of recurrent cholesteatoma had been ruled out and the pathogenesis could be explained due to the presence of the large osteoma arising from the lateral semicircular canal which had gradually obliterated the mastoid ventilation and triggered the chronic infective process within the mastoid cortex leading to fistula formation due to pressure necrosis and wound breakdown over time.

Figure 2 High resolution computed tomography demonstrating the right lateral semicircular canal osteoma arising from the lateral semicircular canal.

OUTCOME AND FOLLOW-UP

The patient was followed up for 6 months postoperatively and she had recovered uneventfully with no recurrence of fistula or discharge at the site of surgery, and normal facial motor function.

DISCUSSION

Osteomas have been reported in all parts of the temporal bone including the squamous, mastoid, middle ear cleft, glenoid fossa, Eustachian tube, styloid process and both internal and external auditory canals.^{1 5 6} Among them, the external auditory canal is the most common location of osteomas in the temporal bone, followed by the mastoid and temporal squama.⁵ Osteomas are composed of well-differentiated, mature bone and characterised by dense lamellae with organised Haversian canals histologically. The intr trabecular stroma contains osteoblasts, fibroblasts and giant cells without haematopoietic cells. Histologically, there are three different subtypes: compact, spongiotic and mixed.^{1 6}

Osteomas of the mastoid cortex or middle ear cleft may remain asymptomatic for a long time before they gradually expand to cause pressure symptoms.^{5 6} Incidental mastoid osteomas are typically managed by monitoring rather than being surgically removed unless causing troublesome symptoms in association with chronic mastoiditis.^{1 2} In case of progressive growth of the osteoma with an increasing hearing loss, ossicular chain involvement, obstruction of Eustachian tube, or round window obstruction, postaural swelling, fistula formation or secondary chronic otitis media, surgery should be considered as treatment of choice.^{5 6}

Ours is an extremely rare case of a large osteoma arising from the lateral semicircular canal obliterating the mastoid ventilation and triggering an infective process within the mastoid leading to fistula formation. Apart from our report, there is only one other reported case of labyrinthine osteoma in the literature from Israel by Ben-Yaakov A *et al* in 2006.⁴ Their patient was also a young woman who had previous ear surgery, which suggests the possibility that it was an acquired osteoma triggered due to trauma or infection similar to our patient. Surgery was the choice in both cases which resolved the symptoms related to the osteoma, with no recurrence in the early follow-up period.

It is prudent for otologists to be aware of this very rare entity since it obscures the normal anatomical landmarks and puts the facial nerve and inner ear at risk during surgery.

Learning points

- ▶ Temporal bone osteoma is an uncommon pathology most commonly at the site of the external auditory canal which can occur at birth or be acquired.
- ▶ Osteoma from the otic capsule is extremely rare but should be considered in patients with predisposing risk factors such as surgery for cholesteatoma or chronic ear disease.
- ▶ Surgery for management of osteoma of the labyrinth is difficult due to distortion of anatomy and needs experienced otology input.

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