

Clinical Records

Middle-ear lipoma as a cause of otomastoiditis

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Abstract

We present a case report which describes a rare cause of a common clinical problem; eustachian tube dysfunction. A seven-year-old child presented with a history of chronic draining ears, despite rigorous medical therapy and multiple ventilation tubes. At myringotomy a mass was noted in the middle ear, and she was taken to the operating room for exploration. The patient was found to have a pedunculated lipoma arising from the anterior medial aspect of the middle-ear cleft producing intermittent obstruction of the eustachian tube orifice. This case represents the fourth case of a middle-ear lipoma in the world literature. We present a review of the literature and an exploration of possible aetiologies of this unusual entity in the differential diagnosis of eustachian tube dysfunction.

Key words: Lipoma; Eustachian tube; Otitis media, suppurative

Introduction

Chronic middle-ear disease due to eustachian tube dysfunction is common. Our understanding of the many clinical manifestations of this process is centred on the concept of a dysfunctional eustachian tube. There are a number of causes of chronic eustachian tube dysfunction. A partial list of these include extrinsic compression (eg. adenoid hypertrophy, nasopharyngeal tumour), inflammation of the eustachian tube mucosa (eg. upper respiratory tract infection, reflux, allergy, cigarette smoke), intrinsic compression (eg. cholesteatoma, polypoid mucosa of the middle ear), or functional obstruction (eg. poor tensor veli palatini function, cleft palate, craniofacial abnormality). Middle ear lipoma is presented as a rarely encountered cause of intrinsic eustachian dysfunction and subsequent otomastoiditis.

Case report

We report a case of a seven-year-old female with chronic eustachian tube dysfunction. She has had bilateral pressure equalization (PE) tube insertions on four separate occasions and an adenoidectomy in the past. At the time of her most recent PE tube insertion, findings clinically suggestive of a right middle-ear cholesteatoma mass were identified. She was then referred to the University of Kansas Medical Center for definitive management.

This child's pertinent medical history in addition to the above includes mild craniofacial abnormalities and developmental delay, with a normal genetic work-up. She suffered from febrile seizures as an infant which have resolved without medication.

Pre-operative audiogram demonstrated pure tone hearing threshold levels of 25 dB on the right for all tested

frequencies, and 5 dB on the left. Examination of the child revealed a tympanic membrane perforation on the left, and a T-tube was present in the right tympanic membrane.

She was taken to the operating theatre and a tympano-mastoidectomy was performed. Operative findings included chronic right mastoid mucosal changes with a poorly pneumatized mastoid bone. She had a near complete mucosal blockage of the aditus ad antrum. In the middle ear, there was a large growth in the anterior medial superior portion of the middle-ear cleft. The mass was well-circumscribed, pedunculated in shape and grossly had the appearance of adipose tissue. The mass almost completely obstructed the eustachian tube orifice (Figure 1). The mass did not appear to encroach upon the ossicles, arose inferior to the eustachian tube orifice and was excised without difficulty.

The gross specimen was a polypoid, tan-yellow soft tissue mass, measuring approximately 3 mm in greatest diameter, with a vaguely lobular pattern on cross-section. Microscopically, the tumour was composed of benign mature fat cells, interpreted as a lipoma (Figure 2). Incidentally noted, there was a scant amount of keratin debris in the specimen, without associated squamous epithelium.

Post-operatively, the child developed a perforation in the tympanoplasty graft due to continued eustachian tube dysfunction. The persistent eustachian tube dysfunction is felt to be multifactorial but definitely complicated by craniofacial abnormalities. Despite a patent T-tube in the right tympanic membrane and the perforation in the left tympanic membrane, two recent audiograms demonstrate bilateral air-bone gaps. A post-operative computed tomography (CT) scan failed to demonstrate evidence of

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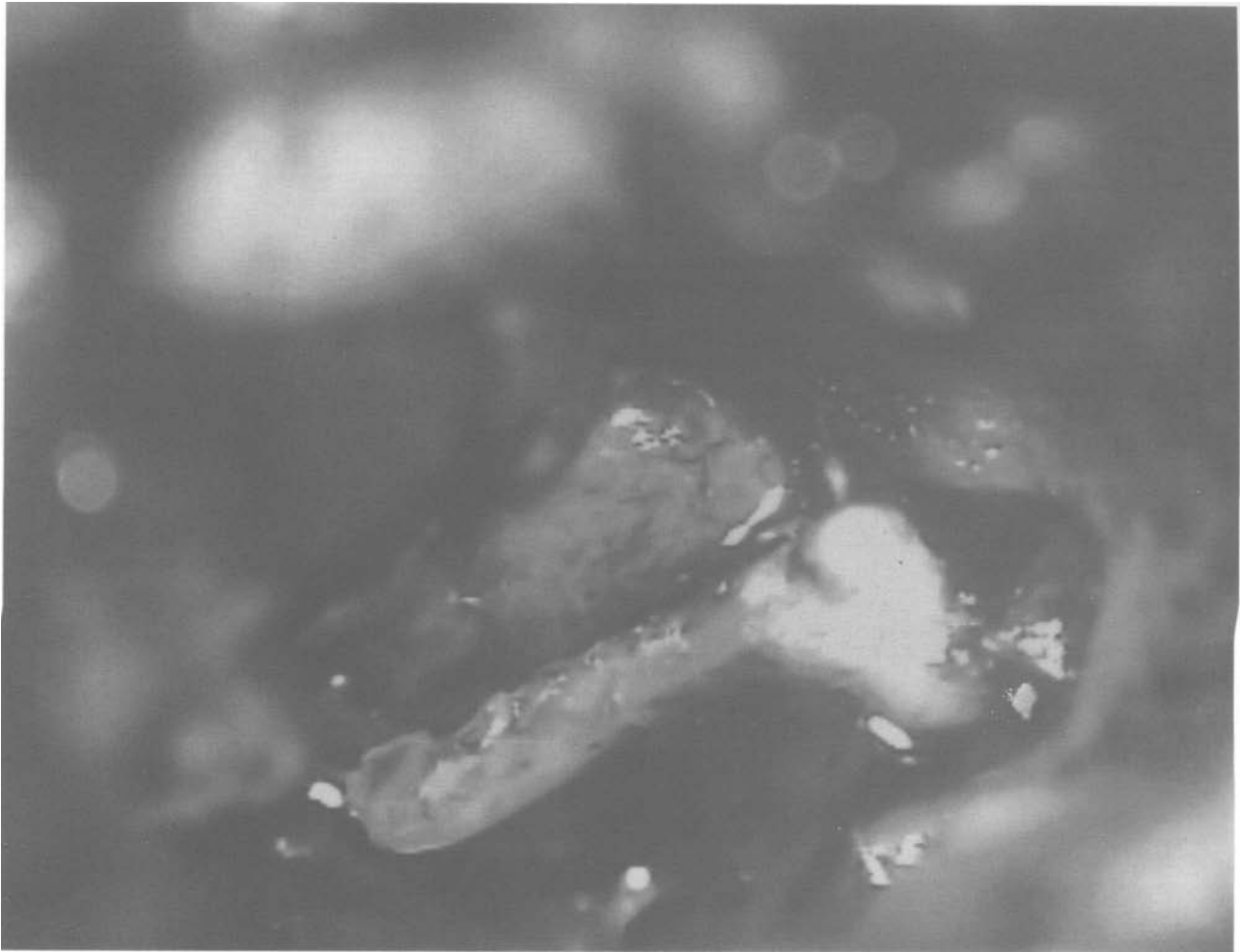


FIG. 1

Middle-ear cleft with tympanic membrane reflected off the malleus. Middle-ear lipoma anterior to the malleus. The mass arose from the anterior promontory region at the mouth of the eustachian tube.

residual lipoma in the right ear nor does it demonstrate contralateral disease.

A follow-up middle-ear endoscopy was recently performed (approximately eight months post-operatively). This was performed primarily to confirm that no cholesteatoma had developed in the post-operative period. The motivation to exclude this possibility was born out of the concern that the keratin debris identified on microscopic examination of the specimen could have come from unrecognized middle-ear cholesteatoma. There was no evidence of cholesteatoma, and the scant keratin debris is assumed to be a contaminant from the external auditory canal. She also had unresolved hearing loss after the initial surgery. An adhesion was located between the umbo and the promontory at the time of the most recent surgery, middle-ear endoscopy. This was ligated and a piece of Gelfilm® (Upjohn) was placed to prevent its recurrence. Follow-up audiogram is pending.

Discussion

The differential diagnosis for a middle-ear mass includes: congenital or acquired cholesteatoma, cholesterol granuloma, inflammatory polyp, facial nerve neuroma, eosinophilic granuloma, adenoma, adenocarcinoma, squamous cell carcinoma, rhabdomyosarcoma, sarcomas, angiosarcoma, or middle-ear lipoma, amongst others. This mass was felt to be a lipoma with a high degree of

confidence intra-operatively, despite its extreme rarity, based on its characteristic appearance.

Adipose tissue is derived from mesenchyme. The principal tumours of this mesenchymal derivative are lipoma and liposarcoma. Lipomas are extremely common benign tumours which are usually asymptomatic and most commonly occur in the subcutaneous tissue of the neck and trunk. Deep-seated lipomas are infrequent and occur in the retroperitoneum and mediastinum. Lipomas usually occur singly but may occur multiply in five per cent of patients and in several lipomatous conditions. Lipomas are composed of mature adipose tissue, and several subtypes occur when other mesenchymal elements are present, such as fibrolipoma, angiolipoma and myelolipoma. Liposarcoma is a fairly common subtype of sarcoma in adults occurring as deep-seated masses predominately in the extremities and retroperitoneum, and fairly uncommonly in the head and neck.

Lipoma and liposarcoma have infrequently been described in the intracranial cavity. A lipoma was first described in the intracranial cavity as an asymptomatic, incidental autopsy finding in 1856 by Rokinsky (Ezinger and Weiss, 1995). Lipomas represent 0.1 per cent of brain tumours, and the most common location for a lipoma in the intracranial cavity is the corpus callosum (Pensak *et al.*, 1986). There have been more than sixteen cases of lipomas described in the cerebellopontine angle (Truwit and Barkovich, 1990). A definitive explanation of the origin

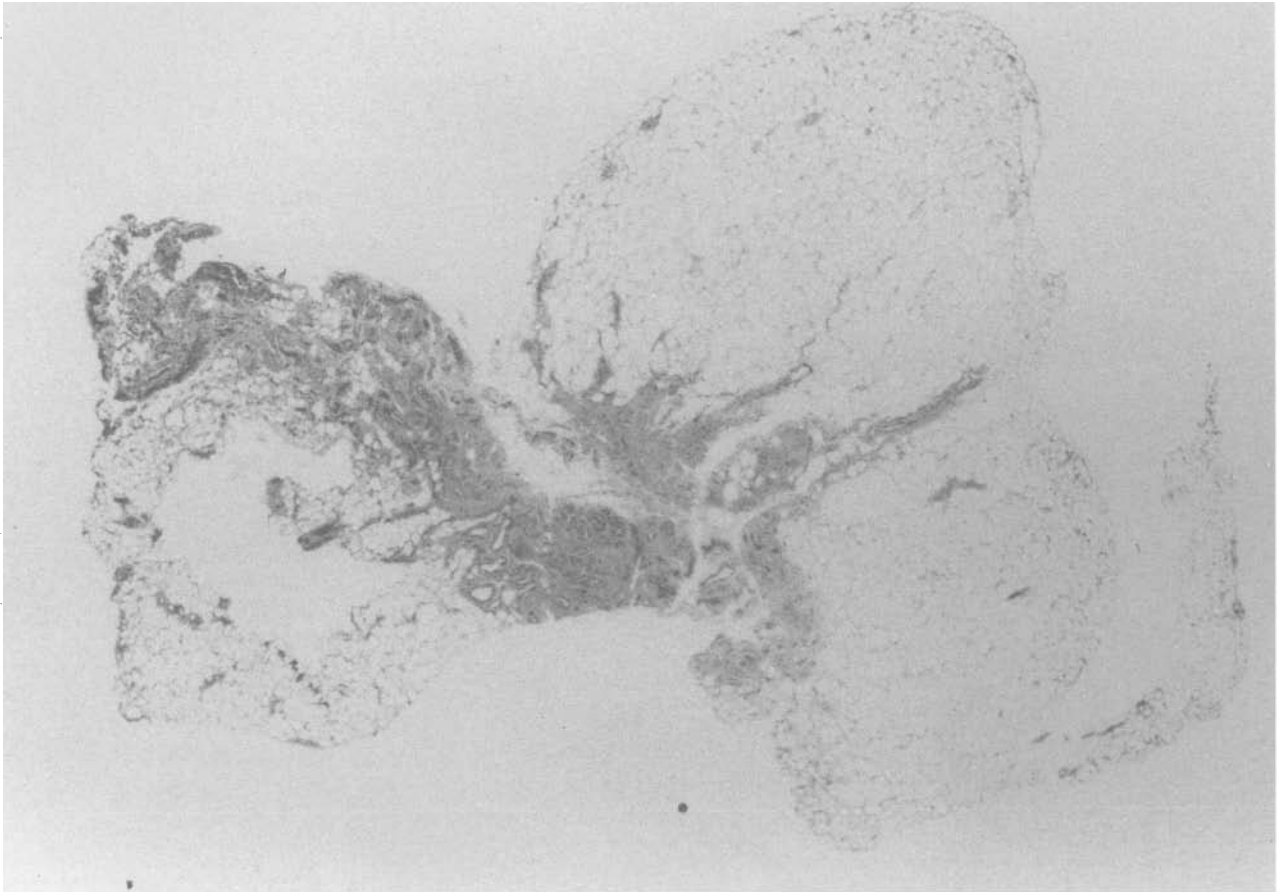


FIG. 2

Low power photomicrograph of the specimen shows a polypoid, lipomatous lesion with a vaguely lobular pattern, composed of mature adipocytes (H & E; $\times 20$).

of the intracranial lipoma is not known. It has been generally described as a result of an abnormal developmental process. In a study of 42 lipomas, over half occurred with other associated brain anomalies, and the authors theorized intracranial lipomas arise from abnormal persistence of the meninx primitiva, which gives rise to the meninges (Youmans, 1982). Others have assigned the probable origin of intracranial lipomas to the pia mater (Olson *et al.*, 1978).

Adipose tumours have been described as arising in the middle ear, although the incidence seems to be even lower than for an intracranial location. The first report of a lipomatous tumour involving the middle ear was of a primary liposarcoma of the mastoid, which produced coalescent mastoiditis, subperiosteal abscess, and facial nerve paralysis (Agarwal *et al.*, 1975).

There have been three previously described middle-ear lipomas in the medical literature. The first case was of a lipoma arising in the middle ear and extending down the eustachian tube, which presented as airway obstruction (Steghuis *et al.*, 1985). In another report, a lipoma was found to arise from the anterior epitympanum and to essentially fill the entire middle ear. The lesion caused conductive hearing loss, which was the presenting complaint. The child reported in that case was reported to have craniofacial abnormalities (Selesnick *et al.*, 1990). The third previously reported case describes bilateral lipomas, again arising from the anterior epitympanum. The patient presented for treatment because of chronic eustachian tube dysfunction (Abdullah *et al.*, 1993).

Thus, the explanation of the pathogenesis of this tumour remains unclear. In multiple anatomical and histopathological studies of the middle ear and eustachian tube, there is a consistent omission of any reference to normally occurring adipose tissue from which these lipomas could have arisen (Buch and Jorgensen, 1964; Akaan-Penttila, 1982; Sade, 1986). There is mention of subepithelial primitive mesenchymal tissue, from which the adipose tissue could have arisen. This sort of proposed aetiology is analogous to the explanation often given for congenital cholesteatoma, that has been postulated to arise from persistent embryonal epithelial rests in the foetal temporal bone (Michaels, 1989). The consistent location of these tumours although few in number does suggest that the cases may share a similar aetiological process.

Summary

Middle-ear lipomas do occur, but are quite rare. Middle-ear lipomas should be considered in the differential diagnosis of a middle-ear mass. Negative pressure in the nasopharynx may draw the lipoma to the eustachian tube orifice, causing intermittent obstruction, and ultimately chronic otomastoiditis.

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