Intrapetrous internal carotid artery aneurysm diagnosed on the basis of middle ear effusion: A case report

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Abstract. The present study reports a rare case of a giant intrapetrous internal carotid aneurysm that compressed the internal jugular vein causing recurrent middle ear effusion in a 13-year-old female. Images obtained by computed tomography revealed middle ear effusion occupying the right side of the attic. Digital subtraction angiography (DSA) resulted in a diagnosis of a giant aneurysm of the right intrapetrous carotid artery, with a diameter of 25 mm and a neighboring area of compression of the internal jugular vein. The patient was treated successfully using coil embolization. The present study therefore indicates that DSA should be considered in the differential diagnosis of patients with middle ear effusion. Early treatment with coil embolization or other surgical treatments can be a life-saving therapeutic approach.

Introduction

Middle ear effusion is commonly observed in otorhinolaryngological practice. Causes of middle ear effusion include viral and bacterial infections, allergies and autoimmune diseases, adenoid hypertrophy, functional abnormality of the Eustachian tube, and gastroesophageal reflux syndrome (1,2). The common symptoms of aneurysm include cochelar nerve damage, tinnitus, hearing impairment, facial nerve and trigeminal nerve palsy. Although computed tomography angiography (CTA) is clinically more commonly utilized and is helpful in the treatment of this condition, digital subtraction angiography (DSA) is the gold standard for diagnosis. The therapeutic results of this combined embolization technique approach have been good, with an 80% survival rate (3). However, the pathogenesis of middle ear effusion remains unclear.

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The present study reports the rare case of a patient who experienced recurrent middle ear effusion as a result of compression of the Eustachian tube by a right-sided intrapetrous carotid aneurysm. When middle ear effusion is the main presenting symptom, as in the present case, the diagnosis may be delayed by the common nature of this complaint. Awareness of this condition is important in order to direct the clinician to perform angiography, which is the definitive investigatory procedure. Once the diagnosis has been established, treatment consists of endovascular coil embolectomy under radiographic control or other surgical treatments with, or without, subsequent reconstruction. Written informed consent was obtained from the patient's family.

Case report

On May 7, 2011, a 13-year-old female presented to Subei Hospital (Yangzhou, China) with a 6-month history of a blocked ear sensation together with hearing loss in the right ear. These symptoms were persistent and marked, but there was no history of vertigo or otorrhea, and the patient had previously been well, with no past medical history of ear disease or trauma. There was no significant personal or family medical history. Upon examination, the right tympanic membrane was dull and indrawn suggestive of a middle ear effusion. The oropharynx showed first-degree enlargement of the tonsils, and electronic nasopharyngoscopy showed adenoid hypertrophy and oropharyngeal mucosal edema. Acoustic immittance revealed a C curve in the left ear and a B curve in the right ear, while pure tone audiometry in the right ear revealed 35-dB conductive hearing loss, with a normal result in the left ear. The provisional diagnosis of right-sided secretory otitis media with adenoid hypertrophy was made on the basis of the previous clinical findings and auxiliary examination. Next, right tympanostomy tube placement and an adenoidectomy were performed under general anaesthesia on the second day of admission to confirm the presence of an effusion and allow ventilation. The post-operative period was uneventful with a significant improvement in hearing. The patient was discharged after three days. The follow-up examinations showed no any sign of recurrence, therefore, the tube was removed after six months.

At 15 days post-tube removal, the patient again felt a sensation of fullness in the right ear and experienced hearing

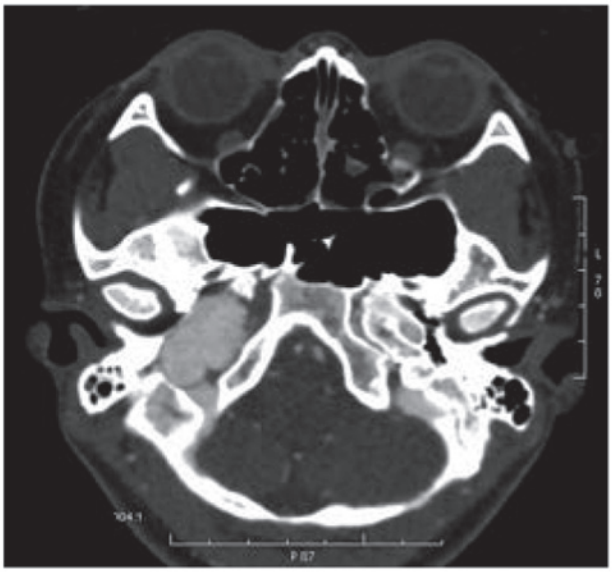

Figure 1. Computed tomography scan of the temporal bone showing middle ear effusion occupying the right side of the attic.

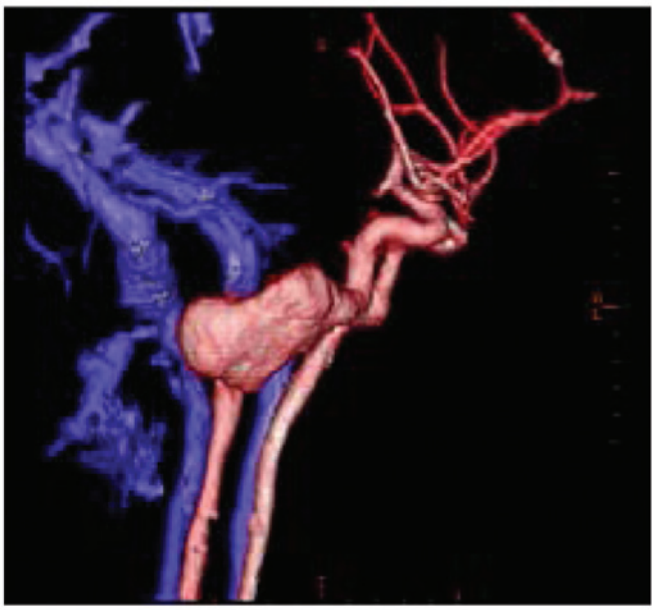

Figure 2. Computed tomography angiography scan showing an aneurysm of the petrous segment of the right internal carotid artery, plus internal jugular vein compression with tortuous dilatation of the tributaries and compression-induced distal convoluted reflux disorder. The largest diameter of the aneurysm was 25 mm.

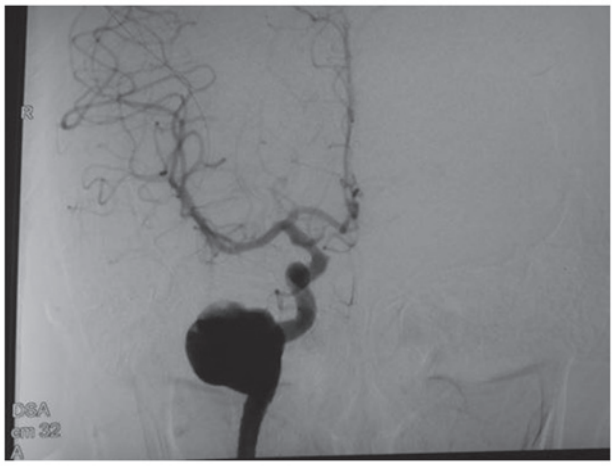

Figure 3. Angiogram of the internal carotid artery. Right internal carotid digital subtraction angiography (arterial phase) showing a 25x16-mm giant internal carotid aneurysm occupying the petrous segment.

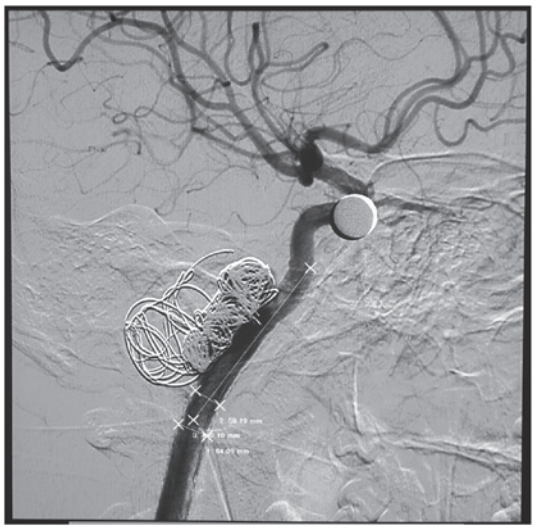

Figure 4. Coil embolization of the right intrapetrous internal carotid artery during right carotid angiography.

impairment. Upon examination, right middle ear effusion was observed. Computed tomography (CT) of the temporal bone again demonstrated middle ear effusion occupying the right side of the attic (Fig. 1). Upon the basis of the clinical findings and the imaging diagnosis, there was a strong suspicion that a giant aneurysm was present in the region of the right internal carotid artery. Accordingly, CTAwas performed, which revealed an aneurysm of the petrous segment of the right internal carotid artery, internal jugular vein compression with tortuous dilatation of the tributaries and compression-induced distal convoluted reflux disorder. The largest diameter of the aneurysm was 25 mm (Fig. 2). DSA was performed via a right femoral approach and an aneurysm of the intrapetrous carotid artery with a size of 25x16 mm was confirmed (Fig. 3). The following therapeutic approaches were considered on the basis of a consultation with the Department of Neurosurgery: i) Embolization of the internal carotid artery aneurysm, performed by placing a coil inside the aneurysm via a catheter; ii) a right anterior craniotomy, followed by clipping of the internal carotid artery aneurysm; iii) a craniotomy followed by trapping of the artery. However, the patient was young and the aneurysm was large in size. In consideration of these and other facts, it was concluded that it would be dangerous to attempt surgery by means of a craniotomy. It was thus decided that the best approach was to perform a coil embolization using a catheter. Initially, the patient and the patients family refused the suggestion of any surgical procedures, including coil embolization, due to the high risks of brain surgery. However, as a spontaneous resolution was

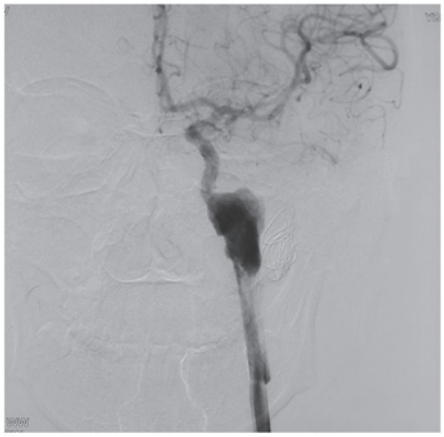

Figure 5. Angiogram showing the significantly smaller post-operative aneurysm and no cerebral insufficiency phenomenon.

not likely, surgery was necessary, and the patient accordingly underwent a successful right internal carotid endovascular coil embolization (Fig. 4). During the procedure, a 6-French guiding catheter (90cm; Boston Scientific, Fremont, CA, USA) was inserted into the right internal carotid artery, a Prowler-14 microcatheter (Cordis Neurovascular, Miami, FL, USA) was guided into the aneurysm and seven orbit coils (Cordis Neurovascular) were released. The patient made a satisfactory post-operative recovery and the aneurysm shrank considerably (Fig. 5). During one year of post-operative follow-up, the patient was completely relieved of the middle ear effusion and the hearing level demonstrated in the pure tone audiogram was preserved.

Discussion

Aneurysms of the internal carotid artery arising within the petrous temporal bone are quite uncommon, and are potentially serious occurrences that are difficult to detect and treat. The causes of the formation of internal carotid artery aneurysms include congenital factors, trauma, mastoid surgery, pharyngeal and tonsillar infection, and corrosion from middle ear disease. Brill *et al* (4) hypothesized that childhood aneurysms can be linked to polycystic kidney disease, collagen vascular disease, subacute bacterial endocarditis, fibromuscular dysplasia, sclerosis, coarctation of the aorta, Marfan's syndrome, syphilis, Ehler's-Danlos syndrome, Moyamoya syndrome, and tuberous and mycotic disease. Such aneurysms are generally congenital in nature (5).

Aneurysms of the internal carotid artery account for all the ~40% of all aneurysms. The most common site of occurrence is the posterior communicating artery, with other sites consisting of the intracavernous internal carotid artery, the ophthalmic artery, the internal carotid artery bifurcation and the anterior choroidal artery (6). The incidence of intrapetrous carotid aneurysms is lower than that of non-traumatic intracranial aneurysms (7,8) Giant aneurysms are more common in children than adults, comprising 31 to 45% of all childhood aneurysms (9,10). An aneurysm size of >25 mm is defined as a giant aneurysm; these occur more commonly in females than in males, and are most commonly found in the connecting section of the cavernous sinus of the internal carotid artery, at the end of the main artery bifurcation, the basilar artery bifurcation and the vertebral basilar artery (11). Aneurysms of the petrous segment of the internal carotid are rare, particularly those with a diameter of >25 mm, and among these, aneurysms with the initial symptom of middle ear effusion are even more of a rarity. A fundamental knowledge of the anatomy of the region is required so that a good understanding of the development of the lesion can be obtained (12). The carotid artery passes vertically through the skull base into the periosteum-lined carotid canal medial to the jugular foramen. The artery lies anteromedial to the tympanic cavity and is separated anteriorly from the Eustachian tube by a thin bony plate. The internal auditory meatus lies posterosuperiorly. The clinical presentation of intrapetrous aneurysms depends on the direction of extension. When the aneurysms extend into the cavernous sinus, the symptoms may include ptosis, diplopia, sixth-nerve paralysis and Horner's syndrome. The symptoms associated with lateral extension of the petrous aneurysm into the tympanic cavity include dizziness, hearing loss, vertigo and pulsatile tinnitus (13). Such aneurysms may remain asymptomatic until their expansion causes mechanical compression of adjacent structures. Aneurysms is this section mainly present with the symptoms of cochlear nerve damage, tinnitus in ~50% of patients, hearing impairment and occasionally, facial and trigeminal nerve palsy. The present study reports a case of a 13-year-old patient in which the history and physical examination did not suggest the symptoms of auditory nerve damage. Therefore, the cause of the aneurysm in the present case was probably congenital in nature. The patient has yet to develop any complications.

Methods for the diagnosis of internal carotid artery aneurysms vary; a combination of CT, CTA, magnetic resonance angiography and DSA may be diagnostic (14). Investigations with CT scans may demonstrate erosion of the petrous temporal bone and effusion may be observed within the middle ear and mastoid air cell system following leakage. In the absence of effusion, such investigations are often unremarkable and clinical judgement must be relied upon to proceed to further analysis. The gold standard diagnostic technique, however, remains as conventional selective carotid angiography prior to therapeutic intervention. Angiography is essential for establishing a final diagnosis and demonstrating the shape, extent and origin of these aneurysms. At present, the use of DSA is becoming more and more common, as it can reveal these lesions accurately with much less risk of complications compared with conventional arterial catheter angiography. In the present case, the patient presented with middle ear effusion as the first symptom, and examination revealed adenoid hypertrophy. Therefore, it was originally believed that the middle ear effusion was caused by adenoid hypertrophy. Thus, tympanostomy tube insertion and an adenoidectomy were performed. The patient relapsed quickly after the removal of the tube, which confirmed that the surgery was ineffective. Next, relevant imaging examinations were performed, which showed an aneurysm of the petrous segment of the right internal carotid artery, temporal bone desorption due to compression and expansive growth to the surrounding, leading to compression of the Eustachian tube and causing tympanic cavity effusion. This case

suggests that in order to form a correct diagnosis, diseases with rare etiology should be considered and the examination should be performed in detail to avoid misdiagnosis and incorrect treatment. Occasionally, middle ear exploratory surgery without pre-operative angiography can result in disastrous results (15). The differential diagnosis of contrast enhancing parasellar masses mimicking an aneurysm in children includes optichypothalamic gliomas, histiocytosis, hamartomas and craniopharyngiomas (16).

Several treatments exist for carotid artery aneurysms, including the following five (3): i) Suturing or clipping of the aneurysm stalk; ii) reinforcement of the arterial wall encompassing the aneurysm by coating it with a synthetic resin adhesive, or by wrapping the aneurysm in fascia or gelatin; iii) intravascular surgical therapy by means of coil embolization; iv) trapping and clipping of the artery on each side of the aneurysm; and v) suturing of the internal carotid artery in the neck region. International studies on unruptured aneurysms have highlighted the fact that the yearly rate of aneurysm rupture is significantly higher in giant aneurysms than smaller aneurysms (17). Thus early interventions should be implemented in cases of giant aneurysms. Direct clipping of the aneurysm is the current gold-standard treatment. If the neck of the aneurysm is wide and cannot be clipped directly then vascular bypass grafting could also be considered (18). In the present patient, due to the broad-based neck and embedding of the aneurysms in the temporal bone, proximal vascular control was difficult to achieve safely (bypass was also difficult) and direct clipping was less likely to be successful. Thus, the best possible treatment approach was spring coil embolization and internal carotid artery ligation. The complication induced by these two methods was an insufficient local cerebral blood supply. Application of internal carotid artery ligatures is the most common method to be applied since 1990, but this has been replaced by coil embolization and other techniques in recent years, placing less burden on the patients (3). Ligation results in thrombosis from the level of the interruption up to the origin of the ophthalmic artery, thereby obliterating the aneurysm. It has been reported that 30-60% of patients treated like this develop neurological deficits and that approximately half of these will succumb (19). Consequently, coil embolization using a catheter was chosen as the method of choice in the present study.

Intrapetrous carotid artery aneurysms occur rarely, and should one present as middle ear effusion, delays in diagnosis may result from the common nature of this complaint. Middle ear effusion in the presence of a haemotympanum should alert the clinician to the possibility of this condition. Whilst CT scans may reveal the lesion, DSA is necessary for a definitive diagnosis and delineation.

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