INTRODUCTION

Infection of the middle ear may be complicated by the spread of infection within the temporal bone into the petrous apex. Petrous apicitis is an extension of infection from the mastoid air cell tract into a pneumatized anterior or posterior petrous apex. In the preantibiotic era, petrous apicitis was more commonly encountered but nowadays it’s very rare. The classic symptoms of petrous apicitis include deep facial pain, constant otorrhea and ipsilateral abducens nerve paralysis. This triad was first described by Giuseppe Gradenigo and later called as Gradenigo’s syndrome.1

This condition is often the result of chronic otitis media with long-standing purulent otorrhea but also may be seen secondary to acute otitis media with or without otorrhea especially in children.2-6 Although traditional management of Gradenigo’s syndrome requires aggressive and radical surgery, there are some reports3-5 which mention the situation less invasively, particularly in pediatric patients. Chronic suppurative otitis media is the main leading cause of Gradenigo’s syndrome in adult population, whereas acute otitis media without otorrhea is more common in pediatric age group.3-5

We report a case of petrositis presenting as the Gradenigo’s syndrome secondary to acute otitis media which was treated successfully by ventilation tube insertion followed by anti-microbial therapy.

CASE REPORT

A 56-year-old man presented with a 3-day history of right eye pain and diplopia especially at right gaze. He also reported that he had pain and fullness sensation in his right ear for 10 days. Oral amoxicillin and clavulanic acid was prescribed by a general practitioner with the diagnosis of acute otitis media. However, his symptoms worsened and hearing loss with constant pain in the right facial and retro-orbital areas has begun 3-days before presentation. On physical examination, the patient
has had right sixth cranial nerve palsy with no other cranial nerve abnormality (Fig.1). Otoscopic examination revealed hyperemia and bulging of tympanic membrane with some fluid in the right middle ear cavity.

In pure tone audiogram there was mild conductive type of hearing loss. Tympanogram was type-B. Clinical findings were compatible with classic triad of petrous apicitis. The diagnosis of the Gradenigo’s syndrome was confirmed by magnetic resonance imaging (MRI) revealing an infiltration of the right petrous apex with a hyperintense signal on T2-weighted images and heterogeneous enhancement of the right petrous bone as far as the apex (Fig.2). The patient underwent a right myringotomy with drainage of serous fluid. Bacteriologic study of the fluid was negative. The hearing loss was totally improved just after the intervention. The pure tone audiograms become normal. Then, intravenous ceftriaxone treatment was started.

After six days of antibiotics, the patient reported significant improvement in his symptoms. He reported complete resolution of his retro orbital pain, but diplopia was persisting especially at right gaze. After 10 days of intravenous ceftriaxone treatment the patient was discharged with instructions to complete a two weeks of oral antibiotics. The patient was followed up after two week intervals and the degree of diplopia and abducens nerve palsy was noted. In the follow-up; measurement of visual acuity and intraocular pressure, retinal and anterior segment evaluation by slit lamp, evaluation of eye movements were done biweekly. Anterior and posterior segment examination by slit lamp, intraocular pressure measurement was normal in all examinations. The visual acuity was 10/10 in all examinations. In the first evaluation of eye movements; the right eye was 30 degree esotropic in straight sight and could not pass the midline in right sight (Fig.1). The degree of esotropia in straight sight was decreased gradually and it was totally resolved in the fifth month. In five months period, diplopia was also gradually resolved. The MRI at sixth month revealed total resolution of the inflammatory process compatible with the clinical findings (Fig.3). After eight months, the ventilation tube was extruded simultaneously from the tympanic membrane and there were no sequellae.

Fig.1: Right abducens nerve palsy, (A) looking straight ahead; (B) looking to the left; (C) looking to the right.

Fig.2: MRI. T2 weighted. Hypersignal of the right mastoid indicated by “arrow” and petrous apex indicated by “*”, (A) axial, (B) coronal.

Fig.3: MRI. Axial (A) and coronal (B) T2 weighted image revealing total resolution of the previous inflammatory process.
DISCUSSION

Gradenigo’s syndrome, first described by Gradenigo in 1907, characterized by constant otorrhea, headache, and diplopia, which is due to inflammation of the petrous apex, is also called petrous apicitis or petrositis.1 Nowadays, with the widespread use of the antibiotics petrous apicitis a very rare condition. The syndrome may be secondary to chronic suppurative infection causing otorrhea or acute petrositis due to acute otitis media without purulent otorrhea. There is no statistical data about the age distribution and the type of onset (acute versus chronic) of Gradenigo’s syndrome. However, considering the literature, it seems that chronic suppurative otitis media is the main leading cause in adult population, whereas acute otitis media without otorrhea is more common in pediatric age group.3-6 On the contrary, our case was an adult patient having acute otitis media presenting with the acute form of a petrous apicitis.

Gradenigo’s syndrome has been mostly treated by radical surgery, but there are some reports managing the situation less invasively. Burston et al. reported two cases of Gradenigo’s syndrome, one in a 70-year-old man and the other in a 6-year-old child, which were managed without the need for radical surgery.6 Marianowski et al reported a case of a 6-year-old child, which was totally cured by myringotomy and antibiotherapy.4

Similarly, Marteau et al achieved total cure in a 4-year-old girl only by antimicrobial treatment.7 The rest of the reported cases were generally treated by radical surgery. In our case, myringotomy with ventilation tube insertion was applied and 10 days of intravenous ceftriaxone treatment followed by oral antibiotherapy. The result was total relief of the symptoms and improvement in the MRI. Al-Ammar also presented a favorable outcome in the treatment of Gradenigo’s syndrome by conservative treatment, but unlike our case, still had recurrent symptoms of the syndrome after the extrusion of the ventilation tube.8

Gradenigo’s syndrome in adults mostly occur secondary to chronic suppurative otitis media characterized by otorrhea. However, in our case it was due to acute otitis media which is the common cause of Gradenigo’s syndrome in pediatric age group. Gradenigo’s syndrome due to acute otitis media is extremely rare in adult population and there is not enough data to describe which treatment modality should be used. In our case, we accomplished total cure by conservative approach.

REFERENCES

1. Gradenigo G. Uber die paralyse des nervus abducens bei otitis. Arch Ohrenheilk 1907;774:149-187. (German)

Authors Contribution:

Burak Ulkumen and Yunus Kaplan confirmed the diagnosis of gradenigo’s syndrome. The modality of treatment was determined by Burak Ulkumen and Yunus Kaplan.