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CASE REPORT

Silent Petrous Apicitis

Oguz Guclu, Ozan Karatag, Hasan Ali Tufan, Sule Kosar, Fevzi Sefa Derekoy

Department of Otorhinolaryngology, Canakkale Onsekiz Mart University Faculty of Medicine (OG, FSD)
Department of Radiology, Canakkale Onsekiz Mart University Faculty of Medicine (OK, SK)
Department of Opthalmology, Canakkale Onsekiz Mart University Faculty of Medicine (HAT)

Gradenigo syndrome is a quite rare complication of otitis media. Typical presentation of this syndrome includes sixth cranial nerve paralysis/paresis, acute or chronic otitis media, and trigeminal neuralgia. In this study, we report on a six-year-old child with incomplete Gradenigo syndrome consisting of petrous apicitis and abducent nerve paralysis without otitis media symptoms and trigeminal neuralgia.

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Introduction

Otitis media (OM) is a common pathology in peadiatric cases. Serious and life threatening complications including petrositis, epidural abscess, subdural abscess, cerebral abscess and meningitis, may be seen with OM. However they become rarely seen nowadays due to the improvement of antimicrobial therapy. Beside this, they may be silent but with dangerous evolution [1]. In 1904, Giuseppe Gradenigo described the syndrome, which carries his name and consists of the following triad: acute and chronic suppuration of the ear; paresis or paralysis of the ipsilateral abducent nerve; and ipsilateral trigeminal neuralgia due to petrous apicitis [2]. The atypical clinical features also need special attention.

The present case report describes the clinical management of a child with incomplete Gardenigo syndrome consisting of petrous apicitis and abducent nerve paralysis without otitis media symptoms and trigeminal neuralgia.

Case Report

During the case management, the current ethics standarts were taken into account.

A 6-year-old boy was admitted into the Ophthalmology Department with sudden onset of diplopia. In his ophthalmologic examination visual acuity was 1.0 and biomicroscopy was normal in both eyes. Inability of lateral deviation of the right eye was noted consistent with right sixth nerve palsy. Dilated

Corresponding address:
Oguz Guclu
Department of Otorhinolaryngology,
Çanakkale Onsekiz Mart University Faculty of Medicine,
17100 Çanakkale Turkey
Tel: 0090 542 2707866
Fax: 0090 286 2180393
Email: droguzguclu@hotmail.com

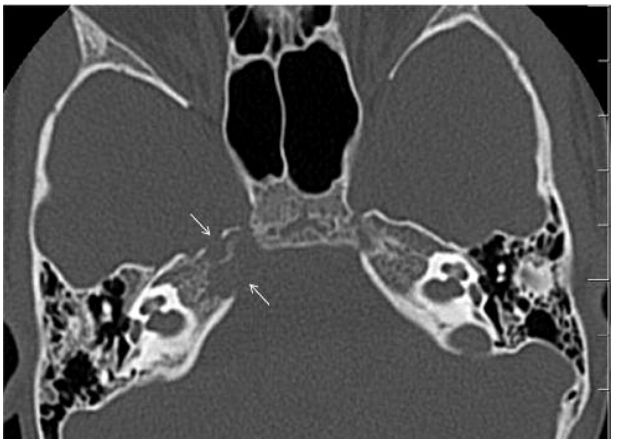

Figure 1. On axial CT image bone expansion and cortical osseous thinning with prominent loss of integrity especially at the proximal of the right petrous apex is seen. Also minimal soft tissue density is visible in some of the mastoid cells on the right.

fundus examination was normal and no afferent pupillary defect was detected. His medical history was unremarkable such as infection, fewer and otorrhea. There was no facial or retrobulbar pain. The rest of physical and neurologic examinations were not unusual. On otoscopic examination, the ear was normal, no effusion in tympanic space. Pure tone audiometry and tympanometry were in a normal range.

The patient's laboratory tests showed a white blood cell count of 7300/mm^3 (reference range 4500-10300) with 49% neutrophil and C-Reactive Protein was 0,25 mg/L (reference range 0-0,8).

A Computed Tomography (CT) revealed bone expansion and cortical osseous thinning with prominent loss of integrity especially at the proximal of the right petrous apex. Minimal soft tissue density was found in some of the mastoid cells (Figure 1). Tympanic cavity was normal on CT examination.

On Magnetic Resonance Imaging (MRI) right petrous apex was found to be minimal heterogeneously izointense to neural parenchyma on T1 weighted (T1W) images and heterogeneously hyperintense on T2 weighted (T2W) images with some expansion. Intravenous Gadolinium enhanced T1W images showed prominent contrast enhancement in the right petrous apex with non-enhancing milimetric hypointense fluid area centrally. There was also mild dural contrast enhancement at the temporal region without evidence of leptomeningeal or parenchymal involvement. These radiological findings were consistent with right petrous apicitis. Also some fluid signal intensities were detected in some of the mastoid cells on the right (Figure 2).

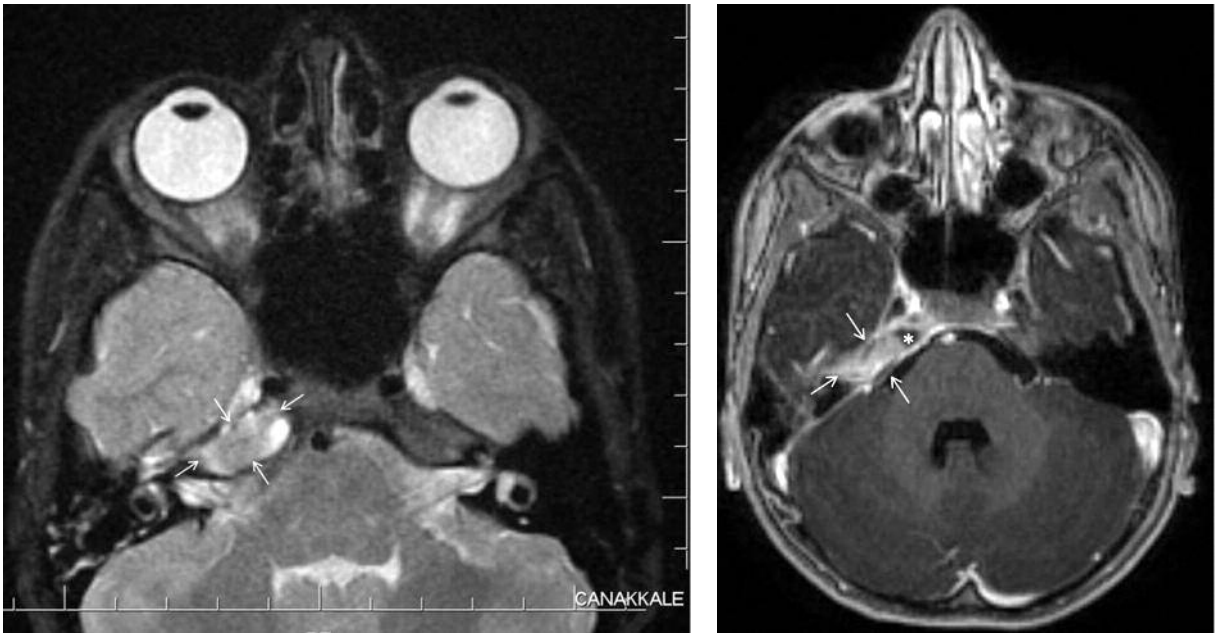

Figure 2. A. T2 weighted axial MRI image shows that the right petrous apex is hyperintense in comparison with contralateral petrous apex with mild expansion (arrows). Some fluid signals are also visible in right mastoid cells. **B.** On gadolinium enhanced axial T1 weighted MRI image, prominent contrast enhancement on the right petrous apex is seen (arrows) inside of which a milimetric non-enhancing hypointense fluid area is also visible (asterix).

According to these findings, the diagnosis of Gradenigo syndrome without OM symptoms was developedconfirmed. He was treated with intravenously administered ceftriaxone (80mg/kg) and metronidazol (400 mg/day). Additionally methyl prednisolon (1 mg/kg) was given for 10 days duration. After two days of treatment, there was a complete recovery of sixth nerve palsy. The patient stayed at the hospital for one-week duration and then the patient was discharged with medical therapy. We stopped medical therapy at the eighth week.

The control MRI examinations obtained 1 month and 2 months following the medical therapy showed that the hyperintensity of the right petrous apex on T2W images and expansion of the lesion was almost completely regressed (Figure 3). Comparing intravenous contrast enhanced T1W images we found that contrast enhancement of the right petrous apex was proceededregressed to some extent on follow-up images with some regression. Mild dural contrast enhancement at temporal region on the first MRI examination was absent on follow-up MRI. Also we detected partial regression of right mastoid fluid intensities on control MRI examinations.

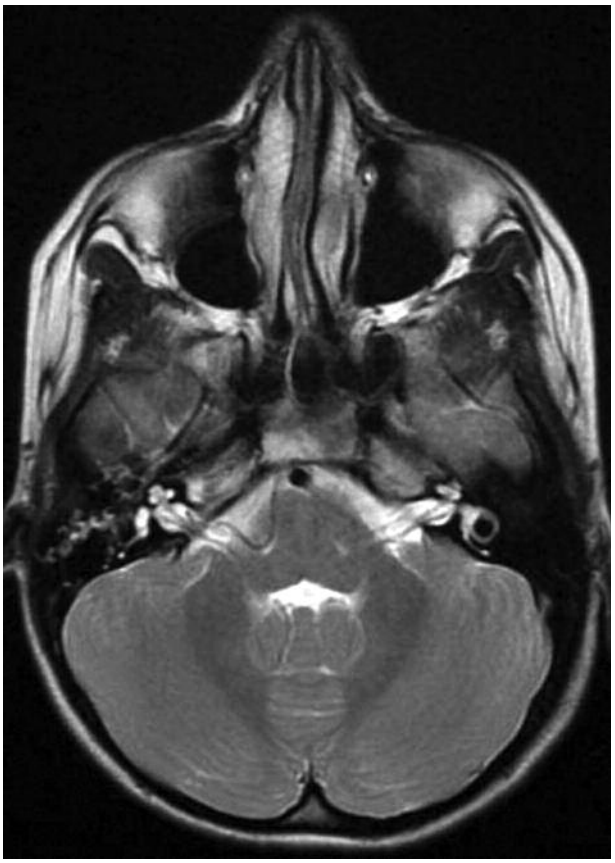

Figure 3. Axial T2 weighted image obtained after 2 months of medication shows prominent regression of the hyperintensity and mild expansion of the right petrous apex lesion. Some fluid signals on the right mastoid cells persist.

Discussion:

Gradenigo syndrome is a rare complication of OM, although major pathology is petrous apicitis (PA). Trigeminal neuralgia and sixth nerve paralysis develop due to PA and OM. Petrous apex is a part of temporal bone and in contact with the cranial nerves, the duramater, vascular structures and the brain. A petrous apex can be undeveloped (sclerotic), can contain marrow, or can exhibit some degree of pneumatization. Pneumatization of the petrous apex develops in only 30% of temporal bones [3]. Petrous apicitis is essentially mastoiditis that occurs in the petrous apex. It is rare because infection in sclerotic or marrow-containing petrous apices is uncommon and the prevalence of pneumatization is low [3]. The trigeminal nerve ganglion and the abducent nerve are separated from petrous apex by only a thin layer of dura mater. In 1905, Primo Dorello described an osteofibrous canal (named Dorello Canal), consisting of abducent nerve and inferior petrosal sinus, at the tip of the pars petrosa temporalis [2]. Abducent nerve is affected in this canal by inflammation [1, 4].

The usual symptoms are diplopia, otorrhea or otalgia and pain, in the region innervated by the first and second division of trigeminal nerve. Facial and retro-orbital pain is secondary to the irritation of trigeminal ganglia and the sixth nerve palsy results in external rectus muscle paralysis and diplopia. Furthermore, deficits of other cranial nerves VII, VIII, XI and X also may be seen with petrous apicitis.

Petrous apicitis without OM symptoms is a very rare condition. We could find only two cases in literature[1, 5]. Actually, the most important thing is that some patients may not have full components of triad and clinicians must be careful. The reason of petrous

apicitis without OM findings can be due to a silent OM entity or long interval between otologic symptoms and the cranial complications. In the present case, the main symptom was diplopia without trigeminal neuralgia or OM findings. Otoscopic and tympanometric examination revealed normal membrane and no fluid in middle ear. However, we found petrous apicitis by radiological investigation. Radiological assessment of petrous apex lesions is needed especially for the extension of the lesions, differential diagnosis and operative planning [6]. Jackler and Parker suggested CT as the first imaging procedure in assessing petrous apex lesions due to high sensitivity and low false-positive rates. They also recommended MRI as a complementary tool to CT [7]. There are several lesions that can involve petrous apex such as cholesterol granuloma, cholesteatoma, effusion, osteomyelitis, and neoplastic lesions [6, 8]. On MRI examination cholesterol granuloma is usually found to have high signal intensity on both T1 and T2 images. Cholesteatoma can rarely involve petrous apex and usually does not enhance on contrast enhanced images. Effusion in petrous apex never presents with bone erosion nor lysis and does not show contrast enhancement. Osteomyelitis in petrous apex and skull base tends to occur in elderly diabetic patients and this entity tends to be more extensive with partial petrous apex involvement. Chordoma can show extension to petrous apex but it is often a midline tumor. Chondrosarcoma can be seen in petrous apex with high signal on T2W images and prominent contrast enhancement. Meningioma and schwannoma are solid lesions that show marked contrast enhancement [6]. Our patient's right petrous apex lesion showed slight T2 hyperintensity with mild contrast enhancement on T1 images and central fluid content. Also there was bone erosion and lysis. Alongside of these radiological findings one important clinical feature was acute presentation of the disease with cranial nerve palsy, which helps in the differential diagnosis of other petrous apex lesions.

Medical and surgical therapy can be preferred according to clinic situation of the patient. Although radical surgical approaches were used in the pre-antibiotic era, recent reports advocate conservative therapy with high dose broad-spectrum antibiotics and less aggressive surgical procedures such as ventilation tubes [9]. The organisms often detected from cultures are group A streptococcus, pneumococus, staphylococcus, pseudomonas aeruginosa and mycobacterium tuberculosis [1, 9]. Ideally, antibiotics should be a choice according to culture; but for initial therapy or culture negative patients, empiric treatment, sensitive to these organisms, can be preferred. Efficiency of steroid therapy is controversial, but it has been used to speed recovery, reduce edema and compression the nerve.

In the recent reports, there is no consensus about medical treatment duration, with a range of 3-5 weeks[1]. Clinical recovery is not enough to end the treatment. We think radiologic investigation is the most helpful argument for not only diagnosis but also for deciding when to stop medical therapy. In the present case, clinical recovery of nerve paralysis is seen at the second day. Maybe this is earlier than in other reports, but we diagnosed the prompt pathology in the fourth day after initial symptoms and started treatment.

As a consequence; Gradenigo syndrome is a rare and serious complication of OM. It can be found in cases without OM symptoms. Radiologic investigation is helpful to diagnose and follow the condition. Furthermore, petrous apicitis must be thought of as an osteomyelitis and followed with a long duration of medical therapy.

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