Case Report

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Radiology Section

A Rare Case of Gradenigo Syndrome: Sequelae to Otomastoiditis

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ABSTRACT

Gradenigo syndrome is a triad comprising of abducens nerve palsy, trigeminal neuralgia and petrous apicitis. It is quite rare complication of otitis media. We hereby report a rare case of a 17-year-old girl who presented with sudden onset of diplopia and left periorbital pain, which on imaging showed otomastoidits, petrous apicitis and epidural abscesses surrounding it.

Keywords: Chronic middle ear infection, Epidural abscess, Petrous apicitis, Trigeminal neuralgia

CASE REPORT

A 17-year-old girl was admitted into the Ophthalmology Department with sudden onset of diplopia and periorbital pain on left side. On examination, her visual acuity was normal. There was inability of lateral deviation of left eye ball indicative of left lateral rectus palsy. Fundoscopy examination was normal. Thus, the probable cause of diplopia was involvement of abducens nerve.

There was no past history of fever. The patient gave old history of left ear discharge. The complete blood count and C-reactive protein count of the patient was normal. After obtaining an informed and written consent from the guardians, a Computed Tomography was performed on the patient.

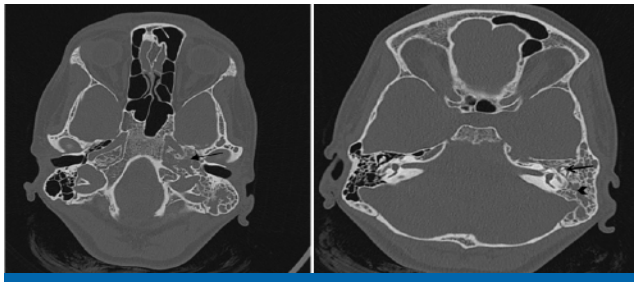
There was evidence of erosion of left petrosal apex and part of left clivus with soft tissue density collection noted in left petrosal apex, left mastoid and left middle ear which is enhancing on contrast administration [Table/Fig-1,2]. Extra-axial biconvex areas of hyperdense collections (HU=89) were noted in left temporal region, left prepontine cistern extending to left CP angle cistern which are enhancing on contrast administration suggestive of epidural abscesses [Table/Fig-3]. The epidural collection in temporal region was measuring about 6 x 8 x 7.5 mm (AP x TR x CC) and the cisternal collection measures about 0.9 x 2.1 x 6 cm (AP x TR x CC) with suspicious extension into left Dorello's canal and Meckel's cave. Ossicular chain was maintained. There was no evidence of erosion of scutum noted. Inner ear structures including cochlea, vestibule and semicircular canals were normal. Internal auditory canal was normal. Facial nerve course and canal appeared normal.

On MRI examination, multiple isointense biconvex collections were noted in petrous part of left temporal bone, left prepontine cistern extending to left CP angle cistern. T2 hyperintensity is noted in left mastoid and left middle ear [Table/Fig-4].

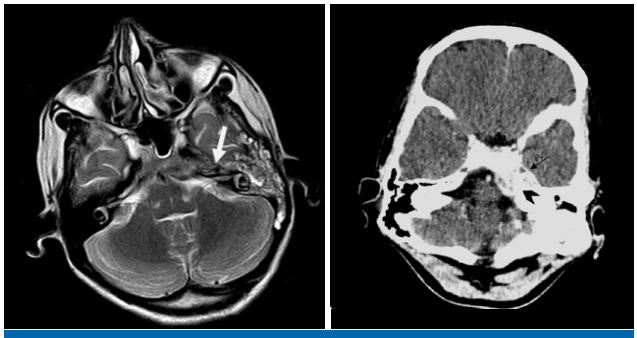
All the findings suggested chronic otomastoiditis with petrous apicitis. Involvement of left Dorello canal and Meckel's cave proved possible involvement of left abducens nerve and trigeminal nerve respectively. Due to involvement of the abducens nerve the patient presented with diplopia. Thus the clinical picture and imaging findings were consistent with Gradenigo syndrome.

A combined medical and surgical approach was advised for this patient. The patient was treated with intravenous antibiotics followed by mastoidectomy and drainage of the involved petrous apex. Post-operative care included regular change of dressings with antibiotic cover for three weeks. The patient responded well to treatment. Steady and progressive recovery of lateral nerve palsy was noted immediately following surgery. After seven days post-operatively it showed partial recovery of lateral rectus palsy on Lancaster test. After thirteen days post-operatively complete recovery of lateral rectus palsy was confirmed on Lancaster test.

Thus, early suspicion of this syndrome, followed by its confirmation on imaging and early surgical intervention aided in good prognosis and total reversal of abducens and trigeminal



[Table/Fig-1]: CT axial image shows erosion and widening of left petrous apex (arrow) and part of left clivus. **[Table/Fig-2]:** CT axial image shows soft tissue density noted within the left middle ear cavity (arrow) and mastoid cavity (arrow head) suggestive of otomastoiditis.



[Table/Fig-3]: CT axial post contrast study shows soft tissue density enhancing collection in left petrosal apex, mastoid and middle ear (arrow head) with extra-axial biconvex enhancing epidural abscesses (arrow) in left temporal region left prepontine cistern extending to left CP angle cistern. **[Table/Fig-4]:** MRI-AXIAL T2 weighted image shows left otomas-toiditis with iso to hyper intense collection in petrous part of left temporal bone, left prepontine cistern and middle ear (arrow).

nerve involvement in our case.

The differential diagnosis for this syndrome includes any neoplastic lesion near petrous apex including sarcoma, meningioma, schwannoma, metastasis or rarely following surgical correction for choanal atresia.

Written informed consent was obtained from our patient for publication of this case report and any accompanying images.

DISCUSSION

Gradenigo's syndrome is a very rare syndrome. It was first described by Guiseppe Gradenigo in 1904 [1]. This syndrome constitutes a clinical triad including otitis media, unilateral pain in regions innervated by the first and second branch of the trigeminal nerve, and ipsilateral abducens nerve paralysis.

Otitis media is a frequent disease in adolescent period and has potential complications in the intratemporal and/or intracranial regions [2]. As mentioned earlier, the incidence of these complications associated with otitis media has decreased since the introduction of antimicrobial treatments. However, their prevalence is still significant [3]. Mastoiditis, facial nerve paralysis, labyrinthitis and acute petrositis are the intratemporal complications of otitis media.

Petrous apicitis have further complications like cranial nerve palsies, meningitis, labyrinthitis, intracranial abscess formation, retropharyngeal abscess and venous sinus thrombosis which may rarely even be fatal [4].There is a communication of air cells in the petrous apex and the middle ear which provide a route for spreading of infections. These include petrositis, petrous apicitis, or otogenic cavernous sinus thrombophlebitis [5].

The spread of the infection from otitis media may also occur by direct extension through facial planes, vascular channels or even bone. Gradenigo syndrome consists of neurological manifestations mainly due to involvement of the fifth and sixth cranial nerves which are separated from the inflamed petrous apex by dura mater [4].

Most of the case of Gradenigo syndrome do not present with all three components [6].

CT and MRI together playing a role of excellent diagnostic tool along with potent antibiotics having good central nervous system penetration has permitted this dramatic change in management of this disease.

CT-images typically show hypodensity with erosive changes at the petrous apex, whereas MRI shows hypointensity on T1-weighted images and hyperintensity on T2-weighted images. Both imaging modalities show prominent enhancement in the area of infection or inflammation.

Sometimes inflammation of middle ear may extend via the tegmen tympani defect and even cause pachymeningitis and MRI is very sensitive in demonstrating these changes.

MRI helps in delineating the differential diagnosis of the many causes of GS which includes cholesteatomas, abscesses, osteomyelitis, neoplasms, and inflammatory granulomas. Studies reported that diffusion-weighted MRI is used to demonstrate abscesses and cholesteatomas in regions of restricted diffusion [7,8].

The most preferred is surgical intervention as the primary management for chronic disease of ear to ensure adequate drainage of petrous apex and mastoid region [5].

Recently, the management of acute petrositis has become less aggressive with methods like mastoidectomies without drainage of the petrous apex combined with myringotomy, tube placement and intravenous antibiotics as compared to previous radical surgeries. Some have even suggested more conservative approach without mastoid surgery. Surgery is still indicated inspite of failure of conservative treatment or chronic ear disease to ensure adequate mastoid and petrous bone drainage [4,5].

CONCLUSION

Our case diagnosed as Gradenigo syndrome associated with

petrous apicitis, trigeminal neuralgia and abducens nerve paralysis as a sequel to chronic otitis media was advised intravenous antibiotics followed by mastoidectomy and drainage of the petrous apex depending on the response to the treatment. Thus, a regular follow-up is required in a case of chronic otitis media and even in absence of classical presentation of Gradenigo syndrome, it should always be kept in differential diagnosis.

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