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Case Report

Diplopia in a Child: A Case of Gradenigo Syndrome

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Abstract

Gradenigo syndrome is a rare but life threatening complication of acute otitis media and/or mastoiditis that needs proper diagnosis and management. Clinically, it is characterized by the triad otitis media, facial pain, and abducens palsy. The majority of cases are secondary to infection requiring long-term antibiotics; refractory cases may require surgical intervention. Knowledge of the etiology and appropriate investigations can facilitate the diagnosis and adequate treatment. We herein present a 4-year-old child diagnosed with otalgia, headache, esotropia, and diplopia. The patient was treated with proper antibiotics and showed good outcomes. This case report highlights the importance of early detection and understanding of the clinical picture associated with Gradenigo syndrome, which have a crucial role in achieving desired outcomes and avoiding serious complications. Gradenigo syndrome should be included in the differential diagnosis of diplopia.

Keywords: *Diplopia; Mastoiditis; Otitis Media; Gradenigo Syndrome; Abducens Nerve, Case Report*

Abbreviations

GS: Gradenigo Syndrome; CT: Computed Tomography; MRI: Magnetic Resonance Imaging

Introduction

Gradenigo syndrome (GS), first described in 1907 by Giuseppe Gradenigo, is characterized by the clinical triad of otitis media, facial pain in the distribution of first and second branch of trigeminal nerve and ipsilateral abducens nerve palsy [1]. It is a serious complication of acute otitis media and mastoiditis, due to the extension of the infectious process to the petrous apex [2]. Cranial nerve dysfunction is caused by osteitis and local leptomeningitis near the petrous apex. In this location, the trigeminal nerve ganglion and the abducens nerve lie closely together, separeted from petrous bone only by dura mater, making them susceptible to any local inflammatory process [2,3]. The full triad of symptoms in GS may not always be present, especially in the post-antibiotic era, but it remains a serious and a life-threatening condition, given their propensity to spread and to involve meninges, cavernous sinus and brain [3].

The correct diagnosis requires imaging studies, namely, computed tomography (CT) scan, magnetic resonance imaging (MRI) or nuclear imaging techniques, to identify the petrous apex as the site of the inflammatory process [3]. The treatment of Gradenigo syndrome is not consensual and should be managed on an individual basis [4]. We report a pediatric case of this syndrome which was managed medically, without the need for sugical intervention.

Case Report

A 4-year-old girl was admitted to our hospital with a 4-day history of right eye pain, headache, nausea, vomiting and diplopia. Twelve days before admission she had presented bilateral otalgia, and amoxicilline and clavulanic acid was prescribed by a general physician for the diagnosis of acute otitis media. However, the symptoms worsened. On admission day, she also complained of diplopia, photophobia and right facial pain. On physical examination, her temperature was 38ºC. Other observations included a heart rate of 92, respiratory rate of 24/min, oxygen saturation of 98% on room air and blood pressure of 114/72 mmHg. Otoscopic examination revealed hyperemia of right tympanic membrane without perforation or discharge of fluid. Ophthalmologic examination showed evident strabysmus and an inability to abduct the right eye (Figure 1). The remainder of extraocular movements were normal. Pupils were equal, round, and reactive to light. There was no afferent pupillary defect. Computed tomography (CT) scan revealed otomastoiditis of the right ear. Brain MRI angiography revealed right acute mastoiditis, petrous apicitis, and partial filling defects of the ipsilateral transverse sinus (Figure 2). Laboratory tests revealed haemoglobin 12.6 gm/dl, White cell count was 6500/mm^3, neutrophils 41% and lymphocytes 47%. D-dimer, C-reactive protein and coagulation factors were normal. There was no growth in blood culture obtained from our patient. A diagnosis of Gradenigo syndrome was made and patient was started on intravenous dexamethasone (0.5 mg/kg/day) for 7 days, and cefepime (100 mg/kg/day), teicoplanin (10 mg/kg/day), and clindamycin (15 mg/kg/day) for 28 days. In addition, the patient received subcutaneous enoxaparin (2 mg/kg/day) for treatment of possible right transverse sinus thrombosis. Improvement was observed in radiological and clinical findings after treatment. Then, the patient was discharged with further recommendations for ophthalmologic examination and was followed up after four week interval. The degree of esotropia was decreased gradually. By the time of 20 weeks clinical follow up examination, diplopia was totally resolved. There were no signs of abducens palsy. Visual acuity was 6/6 in both eyes. Biomicroscopy and ophthalmoscopy were normal in all examinations. During the 5-month follow-up period, no further complications occurred.

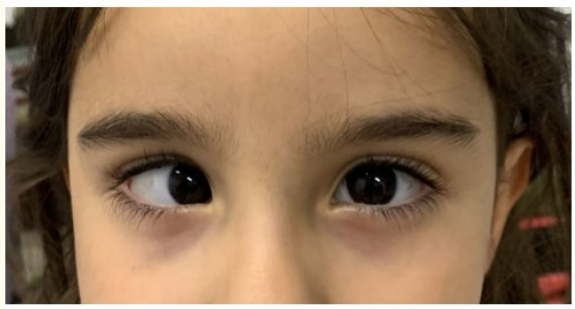

Figure 1: *Patient upon admission with right abducens nerve palsy.*

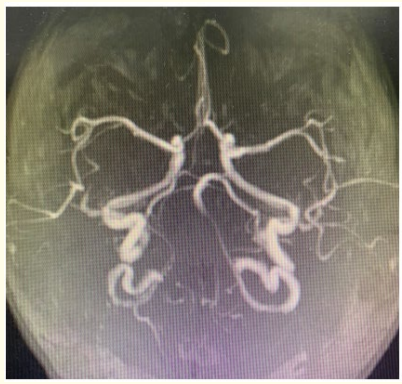

Figure 2: *MRI angiography showing filling defect of the right transverse sinus.*

Discussion

Gradenigo is a rare syndrome that affects about two in every 100,000 children with otitis media [5]. It can, however, be fatal [3,5]. In the modern era of more effective antibiotics, the incidence of intracranial complications secondary to ear infections has substantially declined, with a rate of 2.3% to 6.4% pre-antibiotics and 0.04% to 0.15% post-antibiotics [6]. The local spread of infection to the petrous apex of the temporal bone can lead to meningitis, empyema, brain abscess, venous thrombosis, cranial nerve paralysis and internal carotid hydrocefalus, with a mortality rate estimated in 8 - 26.3% [3,6,7]. The delay between otologic symptoms and cranial nerve involvement may vary from 1 week to 2 - 3 months [2]. In our case, the time between the onset of the initial symptoms and the registration of the abducens palsy was approximately two weeks. This period probably reflected the infection extension into apex.

The difficult clinical diagnosis of GS makes radiologic evaluation essential to petrositis diagnosis, allowing staging and exclusion of other diagnosis, namely septic sinus thrombosis, abscesses, tumours or aneurisms [5-7]. A possible sinus thrombosis would be an alternative explanation for the abducens nerve palsy and diplopia in our patient. Therefore, children presenting with acute otitis media with suspected intracranial complication, should undergo imaging techniques [3,5-7]. CT scan and MRI are helpful tools in the the correct diagnosis and management in most cases. Cerebral MRI angiography confirmed slow blood flow in the right transverse by local filling defects. In this case, based on both clinical characteristics brain image findings, the patient was diagnosed with GS. It is most commonly caused by aerobic microorganisms, but anaerobic microorganisms may also be found [8]. Despite workup no agent was identified in our case.

Therapeutic management of GS remains highly controversial and challenging. Cases as a complication of acute otitis media have usually been successfully treated with high dose broad-spectrum antibiotics. They are recommended and chosen according to culture sensitivity, however, empiriric treatment can be used for initial therapy and culture-negative patients. Radical surgery, usually involving mastoidectomy or petrosectomy, may be required when maximal medical therapy fail, when chronic ear disease is present, or within the presence of potentially fatal complications [2-9].

GS with cerebral venous sinus thrombosis has been rarely reported [10,11]. The transverse sinus is most commonly affected. In this case, a conservative approach was proposed and the patient was managed with high-dose intravenous antibiotics and anticoagulant due to suspection of venous sinus thrombosis. Owing to the absence of otorrea, a culture test of ear fluid was not performed and broad-spectrum antibiotics were administered empirically. Although an optimal treatment duration has not yet been established, Gadre and Chole [10] suggested a treatment period of up to 6 weeks. In our case, the patient was given antibiotics for 4 weeks and improvement was clinically observed. Complete resolution of abducens palsy was obtained within 20 weeks follow-up.

Conclusion

GS is rare but serious complications of otitis media and mastoiditis. Imaging techniques are essential in confirming the diagnosis. In the presente case, the patient received prompt intravenous antibiotics and anticoagulant and achieved a complete recovery without surgery. Colaboration with multiprofissional team involving otorhinolaryngologists, neurosurgeons, pediatricians and ophthalmologists ensure comprehensive care. Early dagnosis and treatment can reduce the complications, leading to significant reduction in mortality.

Conflict of Interest

Authors report no conflict of interest.

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