

A RARE PRESENTATION OF ISOLATED ABDUCENS NERVE PALSY- REVISITING DORELLO-GRADENIGO'S DEBATE

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PRESENTATION OF THE CASE

A 25-year-old male presented with diplopia in our outpatient department. On examination left eye, had esodeviation of 45 prism diopter, limitation of abduction, (Figures 1, 2.) with face turn to left. Visual acuity recorded as 6/6, N6 in both eyes. Fundus examination showed normal fundus in both eyes. Fifteen days before the onset of diplopia, he had been treated for otitis media in other hospital. Vitals were stable and other central nervous system examinations were normal. Otolaryngological examination showed congested left tympanic membrane with air fluid level, absent mastoid tenderness with negative fistula sign and patent eustachian tube. Audiogram showed conductive hearing loss in left ear. Laboratory investigation showed leucocytosis. The magnetic resonance imaging on day of admission was normal. (Figure. 3). Computed tomography of temporal bone also did not show mastoiditis or petrositis (Figure 4).

CLINICAL DIAGNOSIS

Otitis media with inferior petrosal sinus thrombosis.

PATHOLOGICAL DISCUSSION

The abducens nerve is the most frequently acquired cranial ocular motor palsy. Depending on the age of the patient, the common causes of this condition are idiopathic, microangiopathy, cerebral trauma, tumour, demyelination, raised intraocular pressure.¹⁻³ Abducens nerve palsy resulting from direct damage to abducens nerve pathways caused by petrous apicitis (Gradenigo's syndrome), cavernous sinus thrombosis or lateral sinus thrombosis as rare complication of otitis media. Abducens nerve palsy complicated by otitis media without petrous apicitis is very rare, so far only four cases have been documented in literature worldwide.

Our patient on clinical presentation had sixth nerve palsy as initial complication following inadequate management of otitis media without mastoiditis or petrositis clinically (no mastoid tenderness, no trigeminal nerve involvement respectively), as well as radiologically (magnetic resonance imaging, computed tomography of temporal bone

normal) which ruled out osseous spread of infection from middle ear. Therefore, we restricted our diagnosis to vascular spread of infection. Veins of the middle ear drain into the petrous sinuses⁴ Imaging of inferior petrosal sinus was difficult due its small size, hence clinical diagnosis of inferior petrosal sinus phlebitis was made. This diagnosis was further supported by the later involvement of mastoid after 3 weeks of onset of abducent nerve palsy.

Our literature search led us to revisit a historical debate between Anatomist Primo Dorello et al and Otologist Guiseppo Gradenigo et al. Regarding the pathogenesis of sixth nerve palsy in otitis media,⁵ Gradenigo's hypothesis was leptomeningitis both purulent and serous, triggered by the inflammatory process of the cavum tympani.⁶⁻⁸ Dorello after having carried out systematic morphological research on the intra-cranial course of the abducent, stated that he had confirmed the sixth nerve presence at the tip of the pars petrosa temporalis, (not constant) at the inferior petrous sulcus, that was often transformed into a complete canal when the superior wall was closed by a thickening of Gruber's spheno-petrous ligament. This non-extensible canal, within which the inferior petrous sinus and the abducent nerve, were running, represented, according to Dorello, the only area in which the nerve might be compressed by phlogistic post-otitic oedema of the adjacent vascular structures and this compression fully accounted for the relative functional damage.^{9,10,11}

This initiated further research regarding anatomical details of sixth cranial nerve in relation to petrous bone and Dorello's canal. Baldenweck¹² and Baratoux¹³ after their research arrived at the same conclusion as Dorello's. Later Gradenigo also modified his hypothesis of leptomeningitis. Though he agreed that abducent nerve is affected in osteofibrous canal (Dorello's canal) he insisted on osseous route of spread of inflammation from middle ear, while Dorello proposed vascular route that is through veins of middle ear to inferior petrosal sinus, since involvement of abducent nerve following osseous spread of inflammation may lead on to more extensive damage and would leave residual deficit.

We found similar case reports by J. J. Homer et al.¹⁴ He described three patients with acute middle ear infection complicated by sixth nerve palsy without petrositis infection spreading from lateral sinus to inferior petrosal sinus thrombosis. Jung Hyun Jang et al¹⁵ reported similar presentation in a 29-month-old infant with mastoiditis and without petrositis. Out of four reported cases our case has

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findings more in favour of Dorello's conclusion, since the osseous route occurred later in the course of the disease.

This case was reported in order not to lose witnesses to historical debate in our medical field. Also, inferior petrosal sinus thrombosis should be considered as one of the differential diagnosis of acute isolated sixth cranial palsy, since early management would prevent further life-threatening complications such as lateral sinus thrombosis and cavernous sinus thrombosis.



Figure 1. Left Eye showing Esotropia

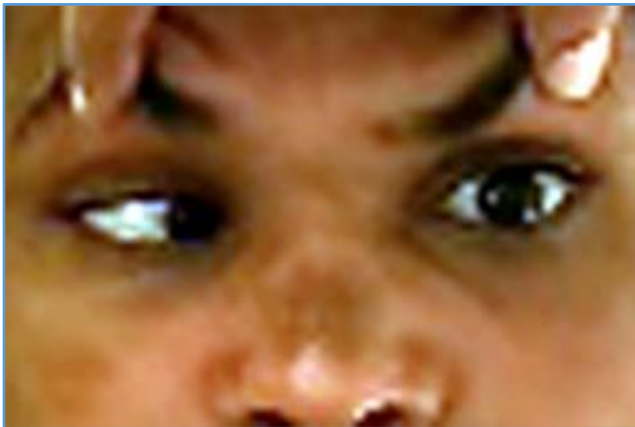


Figure 2. Restriction of Abduction in Left Eye



Figure 3. Normal Magnetic Resonance Imaging on Day of Admission

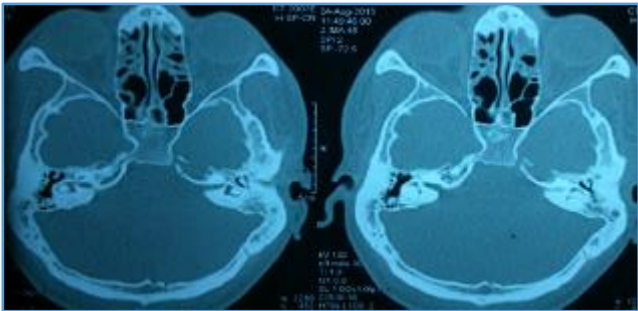


Figure 4. Normal Mastoid and Temporal Bone Computed Tomography on First Admission

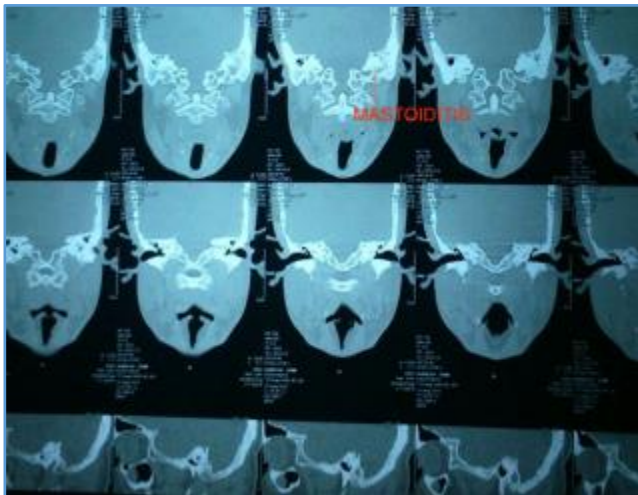


Figure 5. Second Computed Tomography Showing Left Mastoiditis



Figure 6. Left Eye Showing Recovery of Abduction after Treatment



Figure 7. Orthophoric Eyes after Treatment

DISCUSSION OF MANAGEMENT

Patient was started on parenteral ceftriaxone 1 gm twice a day. Patient had improvement in ocular movement within 4 days. He was discharged and advised to continue antibiotics for 2 weeks until review. After 15 days patient again reported with severe earache, retroorbital pain with worsening of diplopia and he had discontinued antibiotics following a week after discharge.

Computed tomography of temporal bone was repeated then showed left mastoiditis (Figure.5). Cortical mastoidectomy was done. Intraoperatively granulation tissue and thick mucopus was noted in mastoid antrum. Patient was treated with parenteral ceftriaxone 1 gram twice a day for one week and Clindamycin 300 mg twice a day for 10 days. Two months after surgery, he recovered completely with full ocular movements and anomalous head posture (Figure. 7, 8)

FINAL DIAGNOSIS

Otitis Media with Inferior Petrosal Sinus Thrombosis and Mastoiditis.

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