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Case report

Successful prolonged conservative treatment of Gradenigo's syndrome in a 4-year-old girl: A case report and literature review

Elodie Marteau [a], Emilie Georget-Bouquinet [a], Suzanne Verlhac [b], Anne Gauthier [c], Natacha Remus [a], Fouad Madhi [a,*]

[a] Service de Pédiatrie, Centre Hospitalier Intercommunal de Créteil, 40 avenue de Verdun, 94000 Créteil, France
[b] Service de Radiologie, Centre Hospitalier Intercommunal de Créteil, 40 avenue de Verdun, 94000 Créteil, France
[c] Service d'ORL, Centre Hospitalier Intercommunal de Créteil, 40 avenue de Verdun, 94000 Créteil, France

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ABSTRACT

Gradenigo's syndrome, one of the complications of middle ear infection, is characterised by persistent otorrhoea, pain in the region innervated by the first and second divisions of the trigeminal nerve and ipsilateral abducens nerve palsy. We report the case of a 4-year-old girl with Gradenigo's syndrome. Computed tomography and magnetic resonance imaging provided evidence of infection of the apex of the petrous temporal bone. The patient received an appropriate antibiotic therapy but the recurrence of symptoms responded to the prolongation of a conservative treatment without surgical intervention.

1. Introduction

The Gradenigo's syndrome (GS), described by Giuseppe Gradenigo in 1907 (also called Gradenigo–Lannois syndrome or petrous apicitis) [1], consists of a triad of purulent otorrhoea, facial neuralgia and paralysis of the sixth cranial nerve. Since the emergence of antibiotics in the systematic treatment of acute otitis media (AOM), this previously well-known complication, has become rare, explaining the difficulties of diagnosis and management today. The diagnosis is based on clinical signs and brain imaging, showing petrositis (osteitis of the apex of the petrous temporal bone due to diffusion of infection of the middle ear via mastoid cells towards its tip). We describe the case of a 4-year-old girl with GS and then we discuss the present management of this syndrome, in particular the indication of anti-microbial therapy.

2. Case

A 4-year-old girl presented with a two-week history of right-sided otorrhea. She received a 6-day course of oral amoxicilline and clavulanic acid and oral prednisolone at a dose of 1 mg/kg/d. The appearance of periorbital facial neuralgia at day 10 and, 2 days later, of paralysis of the sixth cranial nerve suggested the diagnosis of GS. Our patient was afebrile but suffered from headache and vomiting. The leucocyte count was 17 900/mm^3 with a predominance of polymorphnuclear neutrophils (14 200/mm^3) and 2770 lymphocytes/mm^3 lymphocytes. The C-reactive protein was 50.7 mg/l. Bacterial cultures of the auricular secretions, blood and cerebro-spinal fluid were negative.

The diagnosis of the GS was confirmed by computed tomography (CT) and magnetic resonance imaging (MRI) revealing right-sided mastoiditis, involvement of the apex of the petrous temporal bone (Fig. 1), widening and heterogeneity of the lodge of the right cavernous sinus as well as enhancement of the surrounding meninges.

The patient was treated with intravenous ceftriaxone (100 mg/kg/d) and fosfocine (150 mg/kg/d) for 7 days followed by oral amoxicilline and clavulanic acid during 7 days, associated to corticosteroid with prednisolone (2 mg/kg/d) during 8 days. She was discharged from hospital on the 21th day of the disease when facial neuralgia had disappeared but sixth cranial nerve palsy persisted.

On the 32th day, our patient again presented with facial neuralgia of several-day duration and still persisting sixth nerve palsy. The clinical examination found a temperature of 38.1 °C, a change of the general health status (2 kg weight loss in 1 month) with signs of right-sided fifth (V) and sixth cranial nerve (VI) involvement (Fig. 2). There was neither meningeal irritation, nor

* Corresponding author. Tel.: +33 1 45 17 53 98; fax: +33 1 45 17 54 26.
E-mail address: Fouad.Madhi@chicreteil.fr (F. Madhi).

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Fig. 1. MRI. Axial image T2 weighted. Hypersignal of the right mastoid and petrous apex indicated by the arrows.

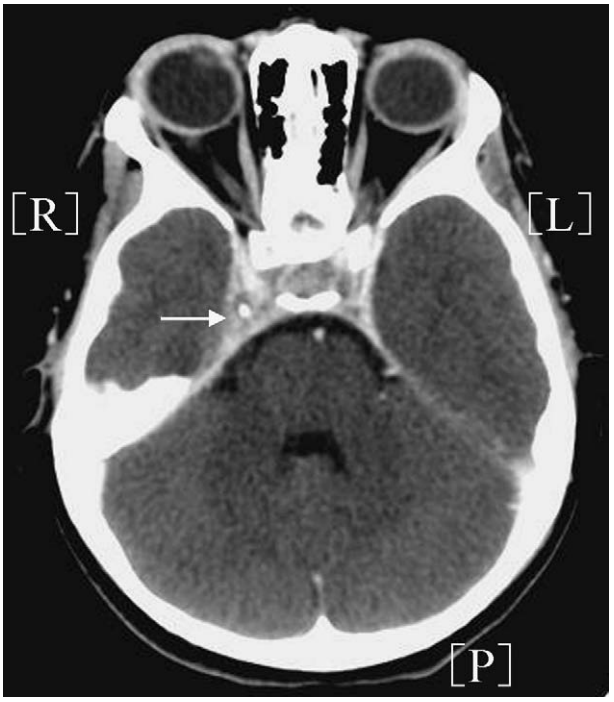

Fig. 3. CT scan. Axial view. Thrombosis of the right cavernous sinus (thickening with a heterogenous aspect like a "comet-tail").

alteration of consciousness. Both tympanic membranes were congestive without otorrhea and the throat was erythematous.

The leucocyte count (9900/mm^3) and the CRP (50.4 mg/l) were only slightly elevated as in the initial presentation and bacterial ear swabs and blood cultures were again negative.

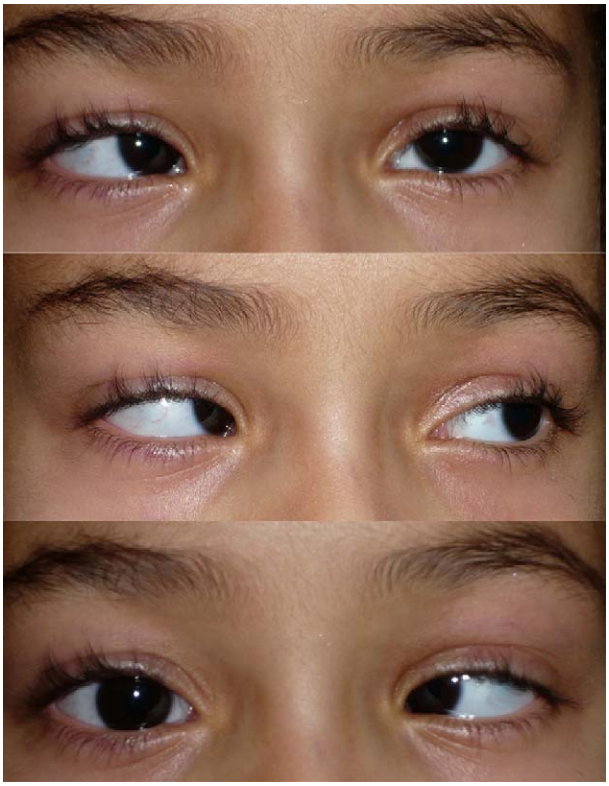

Fig. 2. Right abducens nerve palsy, (A) looking straight ahead; (B) looking to the left; (C) looking to the right.

The cranial CT scan with contrast performed at admission showed right-sided mastoiditis, thrombosis of the right cavernous sinus (thickening with a heterogenous aspect like a "comet-tail") and contrast enhancement of the adjacent meninges without osteolytic lesions or intra-cerebral fluid collection (Fig. 3). The MRI demonstrated a stenosis of the right carotid artery and the precommunicating segment of the right anterior cerebral artery with parietal thickening (Fig. 4).

A triple intravenous antibiotic therapy, including cefotaxime (300 mg/kg/d) and metronidazole (30 mg/kg/d) for 3 weeks and rifampicine (20 mg/kg/d) for 10 days, was followed by a 5-week course of oral amoxicilline and clavulanic acid. A curative anticoagulation with low molecular weight heparin (Innohep 175 UI/kg/d) was introduced for a period of 3 months. The patient was afebrile from the first day of intravenous antibiotic therapy and the CRP became normal after 1 week of treatment. Total immunoglobulin and immunoglobulin subclass levels, lymphocyte

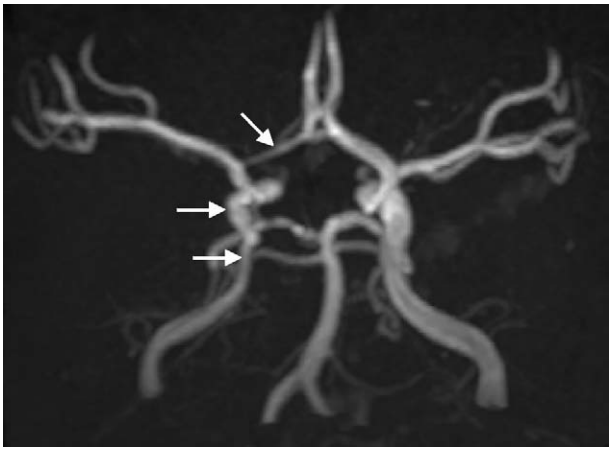

Fig. 4. Three-dimensional time-of-flight (3D TOF) Magnetic resonance angiography (MRA). Willis's polygone. Right carotid and anterior cerebral artery stenoses.