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Case Report

Symptomatic stroke complicating central skull base osteomyelitis following otitis media in a 2-year old boy: Case report and review of the literature

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ABSTRACT

We describe the youngest case to date of a 2 year old child who developed central skull base osteomyelitis (SBO) initially presenting with a fever, vomiting and sore throat. An extremely rare complication of mastoiditis following otitis media in children is SBO which can present with non-specific symptoms. This report describes the first case of symptomatic ischaemic stroke secondary to SBO in an immunocompetent child. We review the literature of the management and the potential cerebrovascular complications of central SBO in children secondary to otolaryngological infection.

1. Introduction

Skull base osteomyelitis (SBO) is rare in children and can present non-specifically as either typical or atypical central SBO. The typical form, well described in the adult population, is predominantly associated with malignant otitis externa in patients with comorbid diabetes mellitus or immunodeficiency. The atypical form, as in our case, is characterised by sphenoid bone osteomyelitis (classically of the clivus), often as a result of infection extending through the Haversian canals from contiguous paranasal infection. It usually occurs in children who are immunocompetent [1].

We describe the first case of symptomatic ischaemic stroke secondary to SBO in a child. Complications of central SBO include cranial neuropathies, sinus thrombosis and abscesses, and it may present alongside Lemierre's syndrome [2]. Symptomatic ischaemic stroke has been documented in combination with Lemierre's syndrome but not in the presence of osteomyelitis in these cases. The arterial cerebrovascular complications of central SBO are usually found as asymptomatic radiological abnormalities [1]. This case highlights the management of central SBO in children and its complications including symptomatic ischaemic stroke, which has previously been undescribed. The literature review examines key radiological and microbiological findings alongside this case to help aid timely diagnosis and facilitate appropriate management.

2. Case report

An immunocompetent 2-year old boy presented with fever, vomiting and sore throat. He was commenced on phenoxymethylpenicillin for presumed bacterial tonsillitis. After 4 days he re-presented with profound neck stiffness, lethargy, mottled skin, tachypnoea and tachycardia. After fluid resuscitation he underwent a full septic screen before being treated empirically with

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ceftriaxone for presumed septicaemia/meningitis. Initial investigations: C-reactive protein 83mg/L; total white blood cell (WBC) count 17.2 $\times$ 10^9/L; neutrophils 8.9 $\times$ 10^9/L; lymphocytes 5.7 $\times$ 10^9/L; cerebrospinal fluid (CSF) WBC count 267/μl (50% neutrophils), CSF glucose 0.3mmol/L (5% of plasma glucose), CSF protein 1.13g/L. Blood and CSF cultures were negative. Computed Tomography (CT) brain scan showed possible periventricular enhancement, internal capsule hyper density and opacification of middle ear clefts consistent with infection. Bilateral otitis media with secondary mastoiditis was diagnosed. Given antibiotics were broad spectrum they were unchanged and surgical intervention was not considered at this stage. The following day he developed a left hemiplegia and left sided homonymous hemianopia. A magnetic resonance imaging (MRI) scan showed signal change consistent with ischaemic infarction of the right frontal lobe [Fig. 1A–B]. There was inflammation/infection of the clivus in the subarachnoid space surrounding the posterior inferior cerebellar artery and MCA, suggesting osteomyelitis, which had not been noted on the original CT scan. Metronidazole, dexamethasone, and aspirin were added to his treatment. He was transferred to a tertiary children's hospital for high dependency care and specialist neurology and infectious diseases opinions. His antibiotics were changed to clindamycin plus meropenem. On day 5 of admission, he had multiple focal seizures (treated with phenytoin) and developed a right sided hemiparesis. Levetiracetam was commenced to prevent further seizures and he subsequently remained seizure-free. A CT angiogram revealed extension of the right MCA territory infarction to involve the cortical grey matter and attenuation of the left terminal internal carotid artery at the location where the MRI had previously shown infective material, presumed to be the likely source of the emboli. Neurosurgical intervention was not deemed beneficial.

Repeat MRI on day 7 confirmed extensive subacute infarction in the right MCA territory and new infarcts in the internal capsule and left frontal lobe white matter [Fig. 1C]. The scan also revealed central SBO secondary to contiguous spread from otomastoiditis, complicated by bilateral cavernous sinus thrombophlebitis and right sided suprasellar empyema. Given this deterioration despite antibiotic and aspirin therapy, clopidogrel was added to maximise prophylaxis against further stroke and he was treated with five days of intravenous immunoglobulin (IVIG) (400mg/kg/day) for a

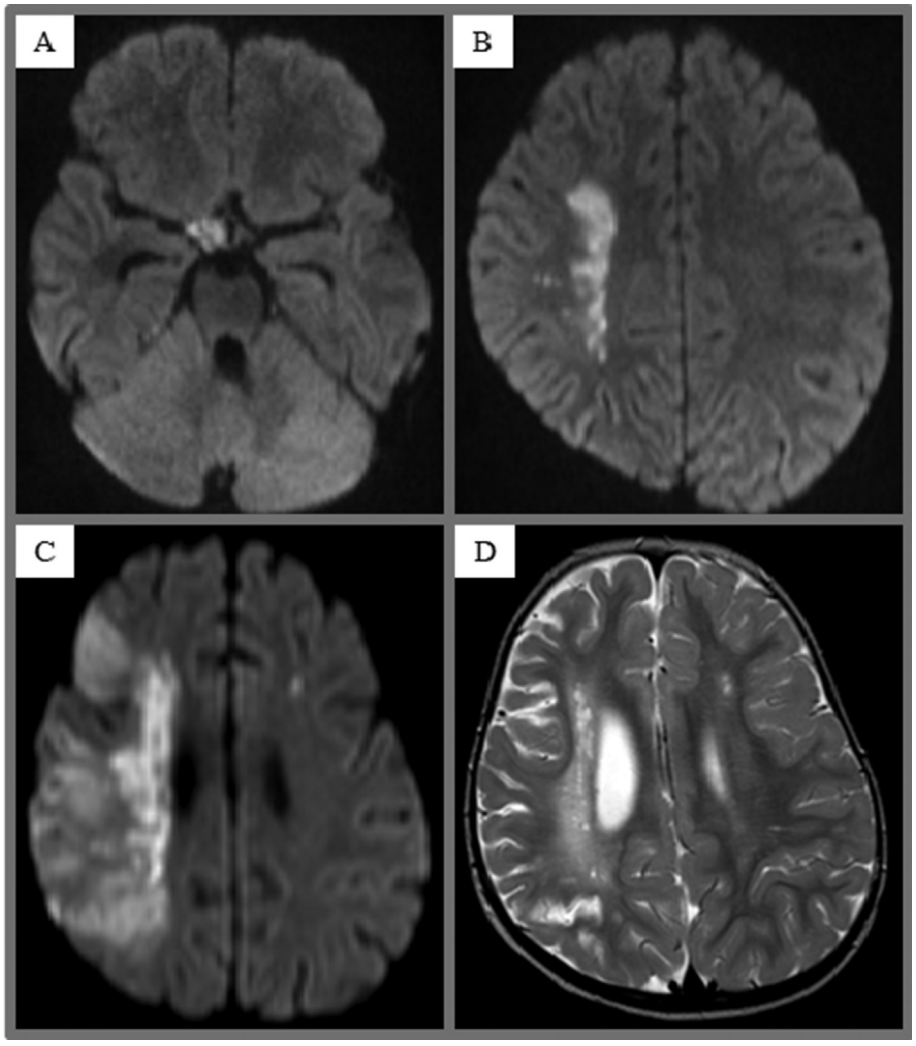

Fig. 1. MRI Imaging of the cerebrovascular complications of skull base osteomyelitis in our case study. A: Initial unenhanced MRI: Axial diffusion weighted images showing material encircling the right supraclinoid internal carotid artery which demonstrates low value on apparent diffusion coefficient (ADC) map. B: Initial MRI: axial diffusion weighted sequences demonstrating areas of acute infarction within the deep white matter of the right frontal lobe, C: Contrast enhanced MRI performed at day 7: Axial b1000 images from a diffusion weighted sequence demonstrating an extensive right middle cerebral artery territory infarct with further new focal infarcts in the left frontal white matter. D: Follow up at 6 months. Axial T2 weighted MRI image showing maturation of the known infarcts, including parenchymal damage with focal areas of gliosis and volume loss involving the MCA territory of the right cerebral hemispheres and to a lesser extent within the left frontal white matter. OUH Foundation Trust Image.