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Otogenic cerebral venous thrombosis in children: A review of 16 consecutive cases



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ABSTRACT

Objectives: Management of otogenic cerebral venous thrombosis (OCVT) is controversial. Despite the modern antibiotic era OCVT still represents a potential life-threatening condition. This study aims to report the clinical presentation and management in a series of children with OCTV. The coexisting intracranial complications (ICC), the extent of the surgical treatment and the role of hypocoagulation were the analysed outcomes.

Material and Methods: Retrospective chart review of patients aged less than 16 years and consecutively treated for OCVT at a tertiary university hospital between January 2007 and March 2015.

Results: Sixteen children with ages ranging between 25 months and 16 years (9 girls/7 boys) with OCVT were identified. Acute otitis media was the causative factor in the majority of cases (n = 13). The remaining cases resulted from chronic otitis media with cholesteatoma (COMC). Eleven patients were under antibiotic therapy prior to admission. Other ICC were simultaneously present: intracranial abscess (n = 6); otitic hydrocephalus (n = 3); and meningitis (n = 1). Thrombus extension correlated with the presence of additional ICC (p = 0.035). Treatment in all cases comprised of broad-spectrum antibiotics, mastoidectomy, and long-range hypocoagulation with warfarin. Transtympanic ventilation tubes were inserted in all cases but one with COMC. Perioperative sigmoid sinus exposure was performed in seven patients, with drainage of perisinus empyema in three cases. Five children underwent simultaneous craniotomy for intracranial abscess drainage. Follow-up imaging performed in 12 cases revealed partial or complete recanalization in three and seven cases, respectively. After a mean hypocoagulation duration of nine months, no hemorrhagic or major neurologic complications were observed. *Conclusions:* The clinical course of OCVT can be masked by previous antibiotic therapy. As such, a high suspicion index is needed for diagnosis. Simultaneous ICC appears to be more frequently found if an extensive thrombosis

was present. The high recanalization rate in this series with low morbidity and no mortality can be obtained with a timely combination of antibiotics, mastoidectomy with transtympanic tube insertion and hypocoagulation. However, the decision to start hypocoagulation and its duration should be undertaken on an individual basis owing the possible adverse effects. Prospective and case-control studies are still needed to better clarify the role of the hypocoagulation treatment in OCVT.

1. Introduction

Otogenic cerebral venous thrombosis (OCVT) is a rare intracranial complication (ICC) of suppurative otitis media [1]. It results either from middle ear or mastoid direct dissemination, or from hematogeneous spread through emissary veins [2]. Although its incidence is low, the potential consequences which may occur in 30% of cases [3] can be serious. Associated ICC include meningitis, intracranial abscess, otitic hydrocephalus and thrombus dissemination to the systemic circulation [4]. Even in the modern antibiotic era OCVT can be fatal from 5 to 10%

of cases [5,6].

The management of OCVT is still controversial. Namely, the extent of the surgical treatment and the role of hypocoagulation are in debate yet.

This study aims to report the clinical presentation and management in a series of children with OCTV, at a tertiary academic hospital. The coexisting ICC, the extent of the surgical treatment and the role of hypocoagulation were analysed.

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2. Materials and methods

The medical charts of children aged less than 16 years with a diagnosis of OCVT consecutively treated at our institution between January 2007 and March 2015 were retrospectively reviewed. Children with otitis media and/or otomastoiditis as a causative condition were selected. The diagnosis was based on otological and neurological evaluation upon admission and complementing radiological findings. All children underwent a computerised tomography scan (CT) during the first 12 h. A magnetic resonance imaging (MR) with gadolinium enhancement was performed if another coexisting ICC was suspected. Medical charts were revised focusing on the clinical presentation, laboratorial parameters, radiological findings, surgical approach, and hypocoagulation treatment. Follow-up was clinical and radiological.

To determine the correlation between the thrombus extension and the presence of coexisting ICC two subgroups were analysed: (A) patients with one cerebral venous sinus affected by OCVT; and (B) patients with more than one sinus affected with or without internal jugular vein (IJV) thrombosis. Statistical analysis was performed with Fischer's exact test with a significance level of p < 0.05 using Prism 6 (GraphPad Software Inc., CA, USA). The study met the approval of the Institutional Board of Medical Ethics (approval process: 11–18).

3. Results

Sixteen children were included in this study, nine girls and seven boys, with age range of 25 months to 16 years (mean age of seven years). Thirteen cases of OCVT resulted from acute otitis media whereas the remainder (patients 10, 15 and 16) were subsequent to chronic otitis media with cholesteatoma (COMC). Mean follow-up was 7.2 years. One case was lost to follow-up. This refers to one child who was transferred to another hospital near the residential area. Eleven patients were under antibiotic therapy prior to admission, mostly amoxycillin/ clavulanic acid. Upon admission, ear pain (63%) was the most frequent symptom, followed by headache (56%), lethargy (44%) and nausea and/or vomiting (38%). Diplopia due to palsy of the abducens nerve was reported in five children (31%). Anorexia and dizziness were present in three cases (19%). Regarding the clinical findings, fever and otorrhea were the most frequent with 11 (69%) and eight (50%) cases, respectively. Spiking fever was common and one child developed



Fig. 2. Magnetic resonance venogram revealing a void sign related to a right sigmoid sinus, transverse sinus and IJV thrombosis (case 2). Please note that intracranial venous circulation is kept by the left side (arrow).

febrile seizures. Besides the aforementioned abducens nerve palsy, papilledema (25%) (Fig. 1) and neck stiffness (19%) were identified in half of cases (n = 8).

On laboratorial analysis upon admission both white blood cell count (between 7470 and 24310 cells/mm³, mean 15514 cells/mm³) and C-reactive protein (mean 132 mg/L) were elevated in the majority of children.

After the first radiological examination, OCVT findings included:



Fig. 1. Limited abdution of both eyes and fundoscopic images showing bilateral papilledema in the same patient (case 2).

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