

Cholesteatoma Complicated by a Cerebral Abscess and Pyoventriculitis: “Spreading Like Wildfire”

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Abstract

Case Report

Cholesteatoma, an abnormal accumulation of keratinized squamous epithelium in the middle ear, is a locally invasive but histologically benign lesion whose silent progression can lead to serious intracranial complications. Although these complications related to chronic cholesteatoma of the middle ear have become rare, they still carry a high mortality rate. This case report presents the rare observation of a cholesteatoma complicated by a brain abscess and pyoventriculitis in a 25-year-old patient, highlighting the crucial role of MRI, particularly diffusion-weighted and FLAIR sequences, in the diagnostic process. Despite multidisciplinary management combining antibiotic therapy and surgery, the outcome was fatal, illustrating the extreme severity of abscess rupture in the ventricular system.

Keywords: Cholesteatoma, Chronic Otitis Media, Otogenic Brain Abscess, Pyoventriculitis, Brain MRI, Diffusion-Weighted Imaging, Intracranial Complications.

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INTRODUCTION

Cholesteatoma, an abnormal accumulation of keratinized squamous epithelium in the middle ear, presents as a locally invasive lesion due to bone erosion, but is histologically benign [1]. Its silent progression can lead to severe complications. Among these, intracranial complications occupy a special place due to their severity and potential mortality, particularly the relatively high mortality rate associated with cerebral abscesses [2]. However, pyogenic ventriculitis, inflammation of the ependyma of the cerebral ventricles, must also be mentioned. This is a rare but serious intracranial complication, often secondary to factors such as cerebral abscesses, and magnetic resonance imaging plays a crucial role in its diagnosis, particularly FLAIR (Fluid Attenuation Inversion Recovery) and diffusion-weighted imaging sequences [3].

The surgical technique depends on the type of complication and the patient's overall condition [4].

In this paper, we present the MRI findings of a case of cholesteatoma complicated by a cerebral abscess and pyoventriculitis. To our knowledge, this is the first publication to combine these three entities.

CASE REPORT

This is a 25-year-old patient with a history of intellectual disability of unexplained etiology. His family had noticed a slight decrease in hearing over the past few months, which had not prompted any further medical investigation. He presented to the emergency department with severe headaches, a high fever of 40°C, and a progressive deterioration of his general condition with altered consciousness. Clinical examination revealed the patient in a state of decerebration with rigidity and pronation of the hands and extension of the legs. Orthoscopic examination was not performed in the emergency department, and the patient was promptly given magnetic resonance imaging (MRI) under sedation.

Brain MRI in this patient showed a left temporal intra-axial lesion with high signal intensity on T2-weighted images, heterogeneous high signal intensity on FLAIR, low signal intensity on T1-weighted images, and diffusion restriction with a central low signal intensity on FLAIR, consistent with a cerebral abscess. Associated with this was content restricting diffusion at the level of the posterior horns of the lateral ventricles suggesting a pyoventricle (Figure 1).

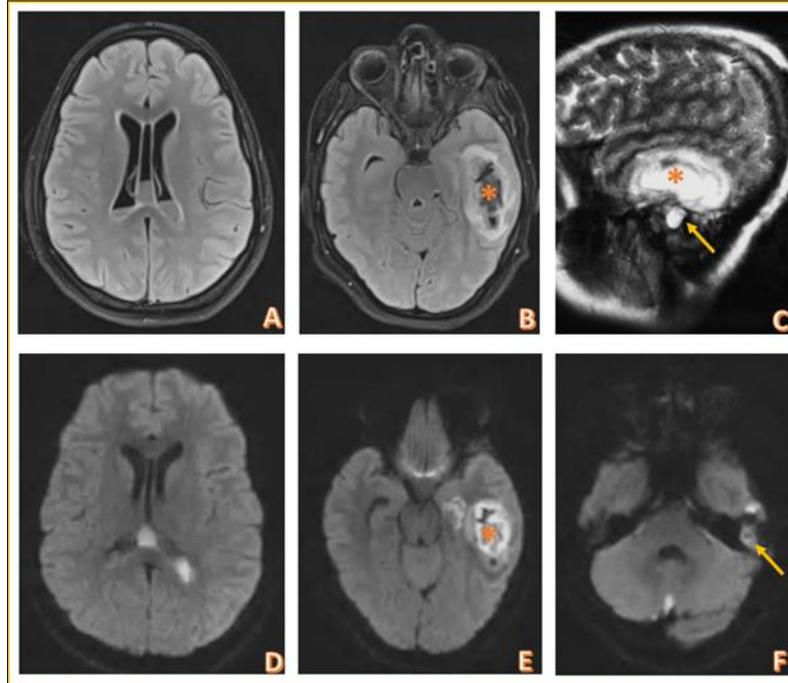


Figure 1: Brain MRI in FLAIR sequence in axial slices (A and B) showing a heterogeneous left temporal lesion process hyperintense at the periphery and hypointense in its center (orange asterisk) associated with dependent filling of hyperintense fluid in the lateral ventricles and the cavum of the septum pellucidum; In sagittal T2 sequence (C) showing the continuity between the process and the filling of the ipsilateral middle ear (orange arrow); In diffusion sequence (b=1000) in axial slices (D, E and F) showing a restriction of water diffusion of the different abnormalities visualized in FLAIR and T2

The condition rapidly deteriorated, with death occurring within hours of the MRI scan, leaving no time for the medical team to intervene surgically.

DISCUSSION

Intracranial complications of chronic cholesteatoma of the middle ear (CCME) are rare today, but potentially serious. Most studies report a high incidence of these complications during the first two decades of life, primarily in male patients [5]. Their incidence has decreased with the advent of antibiotics and improvements in living standards and access to healthcare facilities [6]. In our case, the delay in treatment can be attributed to the patient's intellectual disability, as he did not present with otological symptoms early on.

The clinical picture of cerebral abscess and pyoventriculitis combines an intense infectious syndrome with neurological signs including confusion, altered consciousness, and sometimes meningeal or irritative signs. These generally result from direct spread from a contiguous infected site, such as the sinuses, ears, or mouth; however, hematogenous dissemination or iatrogenic inoculation are also possible [7].

According to most authors, treatment of cerebral cholesteatoma should be initiated early and performed as soon as the patient's neurological condition has stabilized [8].

Cerebral complications secondary to cholesteatoma are, in order of frequency, meningitis, cerebral abscess, and lateral sinus thrombosis [9]. Pyoventriculitis is a dreaded complication of brain abscesses (29%), meningitis (27%), and various neurosurgical procedures (42%), with a hospital mortality rate of 30% and neurological sequelae in 60% of survivors [10].

Complications can be single or multiple. The study by Siba P *et al.*, shows that single intracranial complications account for 56.3% and multiple intracranial complications for 43.7% [11].

MRI is the essential examination for diagnosing ventriculitis and abscesses. In particular, the key sequences, FLAIR and Diffusion, reveal: an intraventricular hyperintensity with diffusion restriction for ventriculitis, and diffusion restriction in the center with a T2 hypointense halo for brain abscesses. The main neuroradiological signs were the presence of intraventricular pus (n = 81, 82.7%), ependymal enhancement (n = 70, 71.4%), and intraventricular septa (n = 15, 15.3%) [10, 12].

The management of intracranial complications of congenital myocardial edema (CME) relies on a multidisciplinary approach combining antibiotic therapy, otological and neurosurgical procedures, as well as adjuvant treatment tailored to specific complications

[11]. The concurrent combination of antibiotic therapy for 4 to 6 weeks, mastoidectomy, and drainage of the cerebral abscess yields remarkable results [3-11]. ENT and neurosurgery can be performed simultaneously, or sometimes the mastoidectomy with cholesteatoma resection is delayed for a few days after neurological stabilization.

Several antibiotic therapy protocols are described in the literature: Combination of a third-generation cephalosporin and metronidazole for 6 weeks, with subsequent adjustments based on the antibiogram [5-10]. More recent studies recommend ceftriaxone combined with vancomycin for broader coverage. Prolonged intravenous antibiotic therapy for 6 to 8 weeks remains the standard, particularly in intracranial abscesses or pyoventriculitis [9-13].

Standard intraventricular neuroendoscopy protocols for the treatment of pyogenic cerebral ventriculitis included one or more of the following interventions: (1) removal of debris, (2) assessment for signs of microbial infection, (3) septotomy, (4) septation incision, or (5) monitoring of catheter placement. A modified external ventricular shunt (mEVS) was associated with enterorespiratory block (ERB) when complete evacuation of intraventricular debris and bacterial plaques was impossible [14].

Mortality from meningitis has decreased dramatically, from 100% before the antibiotic era to less than 10%. It is reported as 0% in the series by Wanna *et al.*, [15], and Raja *et al.*, [16]. A feared complication of brain abscesses is rupture into the ventricular system, leading to pyoventriculitis, which has a mortality rate of approximately 80% [7].

The most frequent neurological sequela is seizures: about 32% of patients who survived 30 days after a brain abscess develop new-onset epilepsy, according to a cohort study conducted from 1982 to 2016 [17].

CONCLUSION

At the end of our work and after reviewing the literature, intracranial complications of cholesteatoma can compromise functional prognosis and, above all, vital prognosis [2]. Imaging plays a crucial role in the diagnosis and monitoring of complications. Diffusion-weighted MRI and FLAIR sequences can be invaluable for detecting intraventricular debris and pus, suggesting ventriculitis. Urgent management by a multidisciplinary team is necessary. However, specific conditions such as intellectual disabilities can mask symptoms and delay intervention before the onset of complications.

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