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Chronic otitis media complicated by mastoiditis and cerebral venous sinus thrombosis in a pediatric patient: a case report

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Abstract

Introduction. Chronic otitis media with cholesteatoma is associated with recurrent purulent discharge and progressive bone destruction and may lead to rare yet serious complications, including mastoiditis, cerebral venous sinus thrombosis and intracranial abscesses. We present a case of chronic otitis media with cholesteatoma diagnosed at an advanced stage and complicated by mastoiditis and intracranial spread.

Case report. A 5-year-old boy was admitted to the hospital with left-sided postauricular pain and mild swelling, purulent ear discharge and episodic headaches. The patient had been treated for otitis externa during the three months prior to hospitalization. Ear examination revealed a thickened, erythematous eardrum covered with purulent discharge. Urgent contrast-enhanced Magnetic resonance imaging demonstrated mastoiditis complicated by the left retroauricular abscess, cerebellar abscess and an extensive thrombosis of the superior sagittal, left sigmoid and transverse sinuses. Emergency tympanostomy, retroauricular abscess incision and drainage were performed. Broad-spectrum antibiotics and anticoagulation were initiated. Following the treatment, purulent discharge persisted daily from the postauricular incision and the middle ear. Two weeks after the initial surgery, a left-sided atticoantromastoidectomy was performed, cholesteatoma and two ossicular bones were removed. One month later, the patient was discharged.

Discussion and conclusions. Post-COVID studies have reported a higher proportion of severe otogenic intracranial complications, including cerebral venous sinus thrombosis, raising discussion about possible associations with hypercoagulability, immune dysregulation and delayed diagnosis. Treatment of cerebral venous sinus thrombosis includes broad-spectrum antibiotic therapy, anticoagulation and surgical intervention in cases associated with mastoiditis, bone destruction, subperiosteal abscess or cholesteatoma.

Keywords: atticoantral otitis; Chronic otitis media; Cerebral venous sinus thrombosis; Mastoiditis.

1. Introduction

Chronic otitis media is a common condition encountered in both children and adults. Based on the type of ear discharge, it is classified into suppurative and non-suppurative (also referred to as serous or mucous otitis media) [1]. The atticointral type of chronic suppurative otitis media is considered the most aggressive one due to its tendency to cause tympanic membrane perforation and destruction of the surrounding bone [2]. This type of otitis media is most associated with cholesteatoma, defined as an abnormal growth of keratinizing stratified squamous epithelium [3]. Cholesteatoma is most frequently located in the middle ear but may also involve the petrous part of the temporal bone or extend intradurally [4].

Chronic otitis with cholesteatoma is considered a relatively rare condition in Europe, with epidemiological studies reporting an overall prevalence of approximately 2.32 cases per 1000 inhabitants [5]. Although cholesteatoma is considered a benign lesion, it is associated with serious health consequences. In children, cholesteatoma tends to follow a more aggressive course compared to adults and is associated with a higher risk of complications [4]. Some authors explain these differences by anatomical factors, such as greater mastoid pneumatization and a higher occurrence of Eustachian tube dysfunction and otitis media in children, which may contribute to disease progression [6]. In addition, molecular studies have shown that pediatric cholesteatoma is associated with increased inflammatory activity and epithelial proliferation, which may contribute to a more aggressive disease course [7]. Complications of cholesteatoma include hearing loss, mastoiditis and potentially life-threatening conditions, such as cerebral venous sinus thrombosis, meningitis and intracranial abscesses [8].

2. Case report

A 5-year-old patient was admitted to the tertiary university clinic complaining of pain and minor swelling in the left postauricular region, purulent discharge from the left ear and episodic but quite intense frontal headaches. A detailed anamnesis revealed a prolonged and worsening condition of ear infection and general symptoms. During the three months prior to hospitalization, the patient had been treated for otitis externa, which had been incorrectly diagnosed. One week before the current admission, mild fever, nausea and vomiting developed in addition to a recurrent ear discharge, leading to a hospitalization in a secondary care hospital. There, the patient was administered intravenous fluids and antibacterial therapy and discharged five days later. Ear examination revealed a thickened, hyperemic tympanic membrane with purulent discharge in the left external auditory canal. The left postauricular region was mildly swollen, with normal overlying skin. Laboratory tests demonstrated leukocytosis and elevated C-reactive protein levels. Ultrasound examination of the soft tissues in the left postauricular region revealed a 20.5 × 9.1 mm cavitory lesion associated with focal sequestration of the bony plate. Given the suspicion of intracranial extension, urgent contrast-enhanced magnetic resonance imaging (MRI) was performed. Imaging revealed left-sided mastoiditis complicated by a left retroauricular abscess, cerebellar abscess and extensive thrombosis of the superior sagittal, left sigmoid and left transverse sinuses.

The patient underwent emergency surgical intervention: left-sided tympanostomy was performed and an incision was made in the left retroauricular region. A large amount of purulent discharge was drained and a rubber drain was placed. No bony destruction was identified intraoperatively at the time. Broad-spectrum antibiotic therapy and anticoagulation with low-

molecular-weight heparin (fraxiparine) were initiated immediately. The patient was admitted to the neurosurgical department. Middle ear culture later grew *Escherichia coli*. Despite treatment, the patient's clinical condition showed only minimal improvement, as headaches persisted and purulent discharge was present daily from both the postauricular incision and the ear canal.

During the second week after the initial surgery, because of the persistent purulent discharge, atticotomy was scheduled. Fraxiparine was discontinued 24 hours prior to the surgery. A postauricular incision was made and mastoid air cells filled with purulent material and granulation tissue were cleared. After widening the opening toward the antrum, atticotomy was performed and a large cholesteatoma extensively involving the tympanic cavity was removed. The incus, head of the malleus and stapes superstructure were removed and the facial nerve was carefully exposed and cleaned. The tympanic membrane defect was reconstructed using auricular cartilage. The wound margins were cauterized to ensure better hemostasis before anticoagulation was restarted and the incision was closed.

Following the second surgery, the patient received conservative treatment in the pediatric otorhinolaryngology ward, including continued anticoagulation, intravenous antibiotic therapy and postoperative wound care. Three weeks later, follow-up contrast-enhanced MRI demonstrated marked radiological improvement with clear retroauricular region and left middle ear, as well as partial regression of the cerebral venous sinus thrombosis. After completion of antibiotic therapy and sustained clinical improvement, the patient was discharged six weeks after the admission to the hospital. Anticoagulation therapy was continued with rivaroxaban and was terminated 6 months later. The patient is periodically observed by an

otorhinolaryngologist, a neurosurgeon and a hematologist.

3. Discussion

This case illustrates a clinically complicated course of chronic otitis media. Initially misdiagnosed as otitis externa, the patient was later hospitalized and operated on for acute suppurative otitis media complicated by mastoiditis, retroauricular subperiosteal abscess, cerebellar abscess and cerebral venous sinus thrombosis. Ultimately the patient was treated surgically the second time and the final diagnosis of chronic otitis media with cholesteatoma was made. The patient underwent a very long path to reach complete recovery. This case report highlights that rare complications of chronic otitis media may occur even in patients without prior illness or immunosuppression.

There are two types of cholesteatoma: congenital and acquired. Congenital cholesteatoma represents up to 5% of all cholesteatoma cases and is believed to originate from embryonic epidermal cell nests [4,9]. It may remain asymptomatic for a long time and is often detected incidentally during otoscopic examination as a white mass behind an intact tympanic membrane [10]. In contrast, acquired cholesteatoma is associated with local infection, chronic Eustachian tube dysfunction and tympanic membrane perforation [9]. Otorrhea is often the first major symptom that leads to the diagnosis of acquired cholesteatoma [10]. If left untreated, cholesteatoma expands and compresses surrounding tissues, while chronic inflammation promotes the production of proteolytic enzymes such as collagenase, acid phosphatase and acidic proteases. These mediators stimulate osteoclast activation, resulting in erosion and destruction of adjacent bony structures [11].

Chronic otitis media with cholesteatoma may be difficult to diagnose, as symptoms can initially be

subtle and otoscopic findings may require examination by an experienced examiner. In chronic suppurative otitis media with cholesteatoma, otoscopic examination may reveal tympanic membrane perforation, purulent otorrhea and visible keratin flakes [1]. To assess the extent of cholesteatoma further radiological evaluation is necessary. Computed tomography (CT) is preferred for the initial evaluation, demonstrating a soft tissue density mass within the middle ear or adjacent structures. CT allows to assess the disease extension, ossicular involvement and potential complications [1]. Magnetic resonance imaging, however, is essential when intracranial complications are suspected or when further characterization of soft tissue is required [4].

Surgery with complete removal of cholesteatoma is the only effective treatment. In uncomplicated cases, an elective surgery is usually planned. Traditionally, surgery is performed under microscopic guidance using a postauricular approach and may include canal wall up mastoidectomy, canaloplasty and posterior tympanotomy to access the tympanic cavity and mastoid region. Although minimally invasive endoscopic cholesteatoma surgery has advanced significantly in recent years, microscopic surgery remains the most reliable and widely used approach in cases where cholesteatoma extends through the aditus ad antrum into the mastoid [9]. In pediatric patients, cartilage grafts are more commonly recommended for tympanic membrane reconstruction, whereas temporalis fascia is more frequently used in adults [4]. In the reported case, the initial surgical management consisted of retroauricular abscess drainage and tympanostomy, as complicated acute suppurative otitis media was suspected. However, slow clinical improvement suggested the possibility of previously undiagnosed chronic middle ear disease. Therefore, an atticoantromastoidectomy was performed, which

resulted in favorable outcomes and noticeable clinical improvement.

Cholesteatoma-related complications are classified as intracranial and extracranial, with extracranial complications further divided into intratemporal and extratemporal. The most common intratemporal complication is conductive hearing loss, which results from the accumulation of cholesteatoma mass, ossicular chain destruction and tympanic membrane retraction, all of which impair sound transmission through the middle ear. As the disease progresses further destruction of the cochlea can lead to permanent sensorineural hearing loss, which is sometimes complicated by tinnitus [12]. Hearing loss is particularly unfavorable in children because of its impact on speech development, social skills and cognitive development [8].

Unlike conductive hearing loss, which is typically a chronic manifestation of cholesteatoma, mastoiditis is an acute intratemporal complication that develops when chronic otitis media is exacerbated by an acute infection spreading to the mastoid. It is the second most frequent intratemporal complication [13]. Extension of acute infection into the mastoid typically presents with retroauricular pain. While uncomplicated cases may be managed conservatively or with tympanostomy, complicated mastoiditis often requires intravenous antibiotics and surgical intervention, including mastoidectomy or, in selected cases, craniotomy [14].

In patients with chronic otitis media with cholesteatoma, untreated acute mastoiditis may promote further spread of infection beyond the temporal bone, increasing the risk of both intracranial and extracranial complications. Extratemporal complications occur when infection extends from the mastoid into the surrounding soft tissues. These complications, such as subperiosteal abscess and, more rarely, Bezold's abscess, require surgical drainage with appropriate antibiotic

treatment [1,4]. Less common extracranial complications include facial nerve paralysis and vertigo [1,13]. Due to slow growth of cholesteatoma, complications often present late. Acute exacerbation may occur with superimposed infection or involvement of critical anatomical structures [8]. In the reported case, the initial presentation mimicked otitis externa, but exacerbation with systemic symptoms (febrile fever, nausea and vomiting, headache) and postauricular swelling raised suspicion of an advanced infectious process. Acute otitis media complicated by acute mastoiditis was the primary diagnosis, however delayed clinical improvement suggested cholesteatoma and necessitated a second surgery.

Intracranial complications of chronic cholesteatomatous otitis media include brain abscess, meningitis, cerebral venous sinus thrombosis, subdural abscess or empyema and otitic hydrocephalus [4]. Although acute and chronic ear infections remain common in the pediatric population, intracranial complications have become rare due to a widespread antibiotic use [15]. However, following the COVID-19 pandemic an increased incidence of otogenic and sinogenic intracranial complications has been reported. Systematic review by Patel et al. reported a rise in cases complicated by cerebral venous sinus thrombosis, parenchymal abscess and meningitis [16]. Some authors have associated this trend with possible post-COVID hypercoagulability, immune dysregulation, and mucosal barrier dysfunction, but the exact mechanism is unclear [17,18]. This increase could also be explained by significant delays in access to routine healthcare during the pandemic and changes in healthcare seeking behavior [16,19].

An association between the patient's age and the type of intracranial complications has been described. A retrospective study by Lee et al. found

that cerebral venous sinus thrombosis, intracranial abscesses and facial nerve paralysis occur more frequently in adolescents and young adults compared to younger children [13]. A retrospective study by Van der Poel et al. suggests that in pediatric patients, symptoms of otogenic intracranial complications more often present with nausea, vomiting, headaches, diplopia – symptoms that mimic meningitis. Whereas adult presentations resemble a stroke – more commonly symptoms include altered mental status, hemiparesis and focal neurological signs [15].

Otogenic cerebral venous sinus thrombosis, on the other hand, is the most common intracranial complication of otomastoiditis in children [14]. It is thought to develop either through direct erosion of the sigmoid sinus bone wall or indirectly through the spread of thrombophlebitis [20]. Magnetic resonance venography is considered the most accurate diagnostic method, although contrast-enhanced computed tomography may also be used [21]. Treatment for cerebral venous sinus thrombosis includes broad-spectrum antibiotic therapy and anticoagulation, whereas surgical intervention should be reserved for selected cases involving mastoiditis, bone destruction, subperiosteal abscess or cholesteatoma [21]. Anticoagulation with low-molecular-weight heparin is recommended during the acute phase and is typically continued for a median duration of 3 months [22]. In clinically stable pediatric patients, maintenance anticoagulant therapy can be changed from low-molecular-weight heparin to rivaroxaban [22,23].

Brain abscess may also be the complication of otomastoiditis and is most commonly located in the ipsilateral temporal lobe or cerebellum and if left untreated, may progress to encephalitis [8]. Typical symptoms include fever, headache, altered state of consciousness and signs of increased intracranial

pressure. In cases of cerebellar abscess, additional neurological signs such as tremor and dysmetria may be observed [8]. Treatment consists of broad-spectrum antibiotics, corticosteroids and anticonvulsants when indicated, as well as surgical drainage of both the abscess and the mastoid [4,8].

4. Conclusions

The reported clinical case highlights the importance of early diagnosis and appropriate management in complicated forms of otitis media. The patient received comprehensive treatment in accordance with current literature recommendations, including prompt surgical drainage of the infectious site, broad-spectrum antibiotic therapy and anticoagulation. Definitive surgical management with atticotomy allowed complete removal of the cholesteatoma and infected tissue, eliminating the primary source of infection.

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