



Management of Luc's Abscess with Extraordinary Clinical Features Resulting in Bilateral Preseptal Cellulitis and Intracranial Complication: A Case Report and Current Literature Review

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Abstract

Luc's abscess is rarely seen complications of otitis media with case reports from the literature despite being known as a benign course. A few cases have been reported combined with chronic suppurative otitis media, unlike other subperiosteal abscesses due to otitis media. Although Luc's abscess is remarkable with its distinctive clinical features, early diagnosis may require high suspicion and sufficient clinical experience of clinicians. In case of clinical doubt, the clinician should not hesitate to request further research. A High-resolution computed tomography scan is necessary for determining the extent of disease and treatment planning as soon as possible. Here we present the case of a twelve-year-old boy with Down syndrome diagnosed with Luc's abscess involving the mastoid bone and developed preseptal cellulitis in both eyes progressed to life-threatening processes. We discussed the clinic features and decision of treatment options of the patients in the light of the literature.

Keywords: Otitis media, complication, preseptal cellulitis, Down syndrome

Introduction

The complication process begins with bone destruction secondary to acute mastoiditis and then spreads the infection to the subperiosteal plane in patients with otitis media. A subperiosteal abscess is called depending on its location (1). Luc's abscess is only encountered in the case reports, even though it was presented as a relatively benign course compared to other subperiosteal abscesses (2,3). Another feature distinguishing Luc's abscess from other complications is the possibility of delay in differential diagnosis due to its rare nature (1). In order to prevent the process of life-threatening complications, early identification is vital. We have experienced that if adequate treatment is not given in time, its aggressive course may be inevitable and behave like a locally aggressive tumor.

This case is presented to guide the routes of infection, show the clinical symptoms that cause a life-threatening process, and emphasize the importance of timely surgical

intervention in possible similar pathologies. In the light of the literature, we wanted to review the clinical approach and surgical planning process in these cases, especially during the pandemic.

Case Report

A twelve-year-old boy with Down syndrome was consulted to our ENT clinic with suspicion of a preliminary diagnosis of sinusitis complication caused by swelling in the right eyelid by the Ophthalmology department. He had a history of purulent, foul-smelling ear discharge from time to time for the last five years. Physical examination revealed inflammatory swelling of the mastoid area with anterior-inferior protrusion of the pinna. The inflammation extended from the temporo-zygomatic region to the right eye, and the examination was painful. The tympanic membrane was not evaluated due to the existing fragile polypoid tissue and edema of the external auditory canal on otoscopic

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examination. Another ENT examination excluding the narrow left external auditory canal was regular.

Laboratory analysis revealed increased C-reactive protein (CRP: 171,4 mg/L; normal<10.0) and leukocytosis (13,8K/uL). Covid PCR tests were done at 24-hour intervals, and they were all negative. Vital signs were within normal limits. There were no pathological findings in the Paranasal sinus computed tomography (CT) imaging.

However, contrast-enhanced high-resolution computed tomography (HRCT) of the temporal bone revealed a few interconnected abscess foci in which the most prominent diameter was 27x18 mm with ring enhancement patterns. Edema and cellulitis extended to the right eyelid and cheek above the temporo-zygomatic region. Both mastoid and middle ear cavities are filled by soft tissue density, and a linear bone defect on the tegmen tympany with existing pathological contrast enhancement was observed (Figure 1). The contrast-enhanced Magnetic Resonance Imaging (MRI) scan showed limited filling defect on the right sigmoid sinus and hypoplastic contralateral sigmoid sinus.

Based on these findings, the patient was hospitalized with the diagnosis of Luc's abscess caused preseptal cellulitis. He was commenced on intravenous (IV) empirical antibiotherapy. In addition, the infectious diseases department suggested adding vancomycin to medical treatment because bilateral preseptal cellulitis occurred with clinical progression despite being treated for three days (Figure 2). The culture of the purulent discharge did not allow a microbiological differential diagnosis. Following clinic regression, a right radical mastoidectomy, repair of CSF leakage, and meatoplasty were performed under general anesthesia after written informed consent was obtained from the patient's parents. During the surgery, we encountered a mastoid bone defect in the Macewen triangle with pus discharge together with widespread destruction of all mastoid cells, middle ear structures, lateral semicircular canal, tympanic segment through the facial nerve's second bend, and the tympanic tegmen caused a visible CSF leak. In addition, the peri-sinusoidal extradural abscess was observed with filled hemorrhagic granulation tissue and cholesteatoma in the mastoid cavity. Following surgery, our patient made an uneventful recovery. Postoperative facial nerve functions were normal. He was discharged with microbiological advice on a two-week oral and topical antibiotics course on the 10th postoperative day. The patient was well and asymptomatic (Figure 3). He completed the 15th month postoperatively with healing and is still under close monitoring and follow-up.

Discussion

Henri Luc, in 1913 first described a subperiosteal abscess spreading to the temporo-zygomatic region



Figure 1. An axial plan CT scan of the temporal bone. Soft tissue density completely filling the right external ear canal, middle ear, mastoid bone, marked edema, the right temporo-zygomatic region (white wide arrow), tegmen defect area (white thin arrow), and defect of the parasigmoidal sinus region (black arrow)

CT: Computed tomography



Figure 2. The right temporal swelling extended anteriorly to the right zygoma and bilateral preseptal cellulitis in a patient with Down syndrome (black arrow). The picture was published with the permission of the patients' parents



Figure 3. In the current image of the patient after 15 months postoperatively, it is seen that there is no sign of temporal swelling and preseptal cellulitis. The picture was published with the permission of the patients' parents

after acute otitis media without mastoid involvement. He claimed that the infection in the middle ear reaches the temporo-zygomatic region through the Rivinus notch from the submucosa of the middle ear or branches of the deep auricular artery. He also argued that the cases of Luc's abscess showed a more benign clinical course since there was no mastoid bone involvement and mastoidectomy was not necessary for their treatment. Although it is already an infrequent clinical entity, we found that case reports supported this theory when we scanned the literature (4). As in our case, Luc abscess cases accompanied by chronic otitis media and mastoid involvement were limited to only a few cases (2,3,5,6). However, we did not find a case report with bilateral preseptal cellulitis accompanied by severe life-threatening complications as a clinical course.

Fernandez et al. (2) reviewed English-language literature to evaluate clinical features and management of Luc's abscess cases from 1989 to 2018. A total of 21 cases (17 children and four adults) were found in the literature. It was observed that only one patient had no signs of mastoiditis in his own CT scan. These patients have been treated with abscess drainage alone, myringotomy with abscess drainage, or mastoidectomy. Although the clinical features of Luc's abscess are relatively stable, it has been emphasized that clinical and radiological evaluation is absolutely necessary before deciding to avoid mastoidectomy, especially in a pediatric patient.

Temporal space abscesses usually originate from an odontogenic infection or acute otitis media. Whereas the most frequent cause of ocular signs is seen in children with an orbital complication of acute sinusitis, the first clinical sign of Luc's abscess may be cellulitis extending from the temporo-zygomatic region to the eye (2,3,7). When examining a child with Down syndrome, difficulties are performing a radiological examination and physical examination due to poor patient cooperation and various structural anomalies such as stenosis of the external auditory (8,9). Therefore, clinical diagnosis can only rely on the physical examination and imaging techniques in

a mentally challenged patient. Unfortunately, our patient with Down syndrome was consulted to our clinic after Luc's abscess had already developed clinically for many possible reasons.

Only 14% of Luc's abscess cases are associated with cholesteatoma (2). We have not encountered such an uncontrolled case of CSOM with cholesteatoma in our routine practice for decades. The conditions of the pandemic period may have caused several challenges to the priorities of both patients and clinicians, even though the only effective treatment for cholesteatoma is still on-time surgery (7,10). We encountered widespread pathological damage caused by cholesteatoma during the surgery in our patient. This case reminds us that CSOM with cholesteatoma behaves like a locally aggressive tumor and should not be considered an elective case, especially during the pandemic.

HRCT temporal bone scan is necessary to confirm the disease's diagnosis and extension, exclude mastoid involvement, intracranial or extracranial complications. It should not be forgotten that extradural abscess is one of the intracranial complications of CSOM that can only be diagnosed during the surgery as seen in our patient (10,11). The necessary surgical interventions should be decided by considering each child's benefit/loss ratio. It was learned that temporal CT was not requested in clinical follow-ups until the consultation was requested from our clinic with a pre-diagnosis of sinusitis complication in our case.

We encountered the child patient with Luc's abscess and an aggressive clinical course. Moreover, this case was perhaps the most advanced form with multiple CSOM complications and bilaterally preseptal cellulitis in the literature. Therefore, clinicians must remain alert for clinical signs and symptoms to examine these patients for the presence of more than one complication. It should be kept in mind that cholesteatoma deprived of early diagnosis and appropriate treatment shows a locally aggressive tumor behavior by its nature.

In conclusion, in case of any doubt during clinical follow-up, evaluating these patients with contrast-enhanced temporal CT may be a vital decision. It should be kept in mind that the clinical course may be more aggressive, especially in comorbid patients who remain uncontrolled with a prolonged suppuration phase.

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Authorship Contributions

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