

Luc's abscess in Down syndrome – A case report

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SUMMARY

Luc's abscess is an exceedingly rare complication of otitis media, where the middle ear infection spreads extra-temporally causing a subperiosteal collection under the temporalis muscle. It is known as a benign complication of otitis media as it is thought not to involve the mastoid bone in comparison to other types of extratemporal abscesses related to otitis media. We describe a challenging case of a 19-year-old male with Down syndrome diagnosed with Luc's abscess involving the mastoid bone. A high-resolution computed tomography scan is important to determine the extent of the abscess, with or without mastoid involvement, and the presence of complications. These findings will then help to determine the surgical options. Drainage of abscesses is a simple, initial, and conservative approach but less effective compared to mastoidectomy. 'Mastoid-sparing' approach should only be considered if there is complete resolution after a simple drainage and antibiotic treatment.

INTRODUCTION

Otitis media (OM) is a very common entity in childhood and is usually self-limiting and rarely progress to complications due to advancement in modern antibiotics.¹ However, complications are on the rise due to growing antibiotic resistance and increasing number of immunocompromised patients leading to significant morbidity and mortality.¹ The incidence rate in developed countries range from 1.2 to 3.8 per 100,000 persons.² Complications of OM are well known to be categorized into extracranial and intracranial complications. Subperiosteal abscesses are rare extracranial and extratemporal complications of OM and are defined by location. *Bezold's* abscess is located deep to sternocleidomastoid muscle, while *Citelli's* abscess is situated in digastric triangle. For Luc's abscess, the infection is located deep to the temporalis muscle.³ Due to its rarity, complications maybe missed due to lack of experience of clinicians who treat this disease.

Luc's abscess is different from other types of subperiosteal abscess as infection can spread from the middle ear through an anatomical pathway in the ear canal without erosion of the cortical bone. This was the description by Henri Luc in which subperiosteal temporal abscess is due to otitic origin without intraosseous destruction.⁴ However, our case below was not in accordance with this theory as there was involvement of the mastoid bone.

CASE REPORT

A 19-year-old man with Down syndrome presented with intermittent right otorrhea for 1 month duration which was purulent in nature, and it was associated with ear itchiness. The history was solely dependent from the mother who is the caretaker. She claimed he never had any history of ear discharge prior to the current presentation and the ear discharge resolved after completing 1-week course of oral Augmentin. However, 2 weeks post-treatment, he started to develop right temporal swelling, which increased progressively in size spreading to the post-auricular region then to right cheek region associated with right mucopurulent ear discharge. The swelling was also painful. His mother denied that the patient had fever, symptoms of rhinitis, headache, or history of trauma.

On examination, there was a diffuse swelling at the right temporal, superior half of post-auricular and zygomatic region which were tense, tender, and erythematous (Figure 1) pushing the right pinna anteriorly. The right cheek and lower eyelid also had mild edema. Otoendoscopy showed an edematous right external ear canal (EAC) with prominent sagging from superior wall without visualization of the tympanic membrane. The left EAC was narrow and dry with an intact tympanic membrane. In addition, the facial nerve function was intact bilaterally, and other nose, throat, and neck examinations were unremarkable.

Laboratory analysis revealed increased C-reactive protein (CRP: 47.0 mg/L; normal <10.0) and slight leukocytosis (12.89 K/uL). Pure tone audiogram as a baseline hearing level demonstrated a moderate to severe hearing loss on the right side and a mild hearing loss on the left side with a type B tympanogram bilaterally.

High resolution computed tomography (HRCT) of the temporal bone revealed a hypodense collection in the right temporal region measuring 6.1 cm (AP) x 1.7 cm (W) x 4.7 cm (CC) with ring enhancement pattern (Figure 2). Both mastoid cavities were sclerotic with soft tissue density in both middle ear cavities. There was erosion of the right mastoid cortex, right incus, stapes, scutum, right tegmen tympani and lateral wall of tympanic part of the facial nerve. Subsequently, the patient underwent examination under anaesthesia, incision, and drainage of the right temporal abscess through Wilde's post aural incision in which 20cc of pus was drained and sent for cultures and acid-fast bacilli (AFB) smear. The right EAC was cleansed and packed with

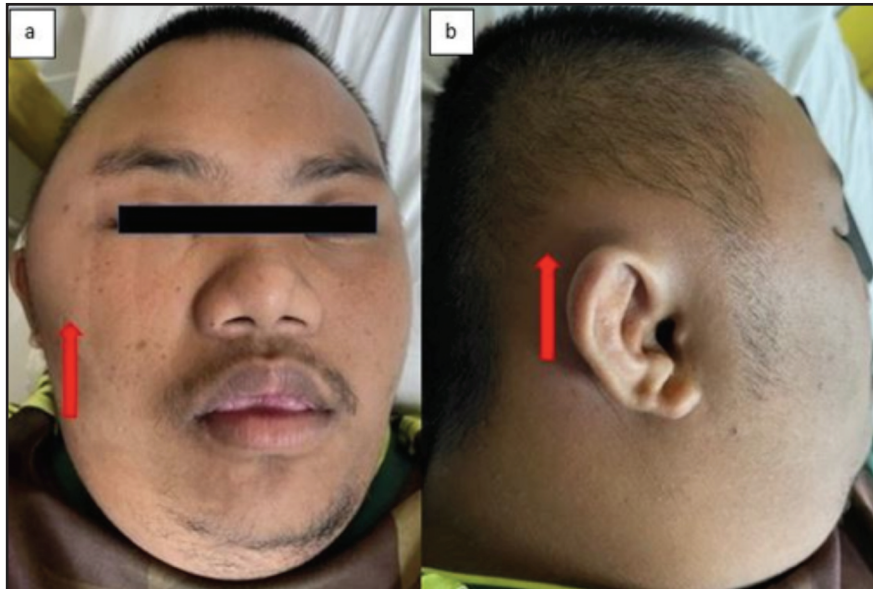


Fig. 1: (a) The right temporal swelling extended anteriorly to right zygoma and lower eye lid (red arrow) and (b) posteriorly to post auricular region (red arrow).

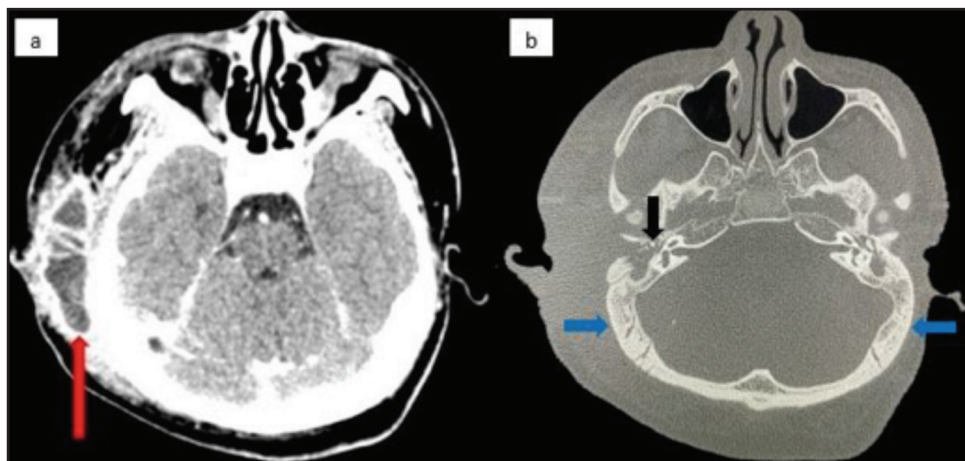


Fig. 2: (a) Heterogenous collection in the right temporal region with ring enhancement in axial soft tissue setting (red arrow). (b) Bilateral sclerotic mastoid air cells (blue arrow) with soft tissue density and erosion of right ossicles (black arrow).

otowick soaked with anti-microbial solutions. Excessive bleeding was encountered intraoperative which made visualization and removal of disease in mastoid and middle ear difficult. Hence, combined approach tympanoplasty (CAT) which was the definitive plan to address the condition of the mastoid and middle ear was planned later after the swelling and oedema subsides. He was initially started on empirical broad spectrum intravenous antibiotics and was later changed to Piperacillin-tazobactam 4.5g tds for one week according to cultures sensitivity which grew *Pseudomonas aeruginosa* and *Streptococcus viridans*. He was discharged after 3 weeks of admission with oral Ampicillin-sulbactam 375mg BD for one week after marked improvement of the temporal swelling and a healing post auricular wound. During subsequent follow up, the patient was well and asymptomatic, however, the parent refused further surgical intervention. This patient is still under our close monitoring and follow up.

DISCUSSION

Henri Luc in 1913 first described the subperiosteal temporal abscess related to OM without involvement of the mastoid bone. He suggested the possible route of bacterial spread is from the submucosa of the middle ear through the notch of Rivinus, deep auricular artery branches towards the subperiosteal area.⁴ Therefore, Luc believed mastoidectomy is not needed as there were absence of clinical signs of mastoiditis such as post auricular swelling and persistent otorrhea. This theory was also supported by Weiss et al in 2010.⁵ However, our case differs from the original report by Luc, as the abscess developed as a complication of mastoiditis with erosion of its cortex.

Luc's theory has been brought into question by an increasing number of cases for both children and adults presented with a subperiosteal abscess underneath the temporal muscle with concurrent mastoiditis. This has been proven by a systematic

review on the clinical features and managements of Luc's abscess based on case reports collected from 1989 to 2018, with a total of 21 patients (17 children and 4 adults).⁶ It reported that almost all patients (95.2%) except one showed signs of mastoiditis in computed tomography (CT) scan,⁶ hence not in accordance with Luc's theory.

The commonest presenting symptoms of Luc's abscess were otalgia and swelling at zygomatic region followed by fever and malaise especially in children.⁶ The site of swelling may involve multiple sites such as the preauricular, temporal, cheek, eyelids, and mastoid area in descending order of frequency.⁶ Our patient presented with otalgia and temporozygomatic swelling without fever. Diagnosing Luc's abscess could be a challenge especially in our case as the patient was a poor historian. Signs and symptoms could be masked or misjudged by clinicians in view of his underlying Down syndrome. Thus, clinical diagnosis can only be relied on the physical examination and imaging techniques in a mentally challenged patient. Furthermore, Luc's abscess could easily be misdiagnosed as orbital complications of acute rhinosinusitis. This is due to frequent association of zygomatic, eyelid and cheek swelling with rhinitis symptoms.⁶ Mastoid swelling with anterior displacement of pinna usually developed few days after preauricular swelling.⁶ Fortunately, otalgia being the commonest symptoms could pinpoint to the otic origin to aid our diagnosis. Only 14% of cases are associated with cholesteatoma.⁶ Risk factors for developing Luc's abscess are drug users and diabetic individuals, but the majority of cases are without risk factors, similar to our case.⁶ Our patient could have been a case of undiagnosed chronic inactive OM evidenced by the sclerotic mastoid air cells. Furthermore, Down syndrome patients has been proven to have an intrinsic defect of their immune system leading to higher frequency of infections and complications.⁷

HRCT Temporal bone is the imaging of choice, and we recommend this being done promptly. This is to exclude intracranial and extracranial complications, confirming diagnosis, disease extension, and status of the mastoid. HRCT scan could aid the decision making of either doing mastoidectomy or local drainage only. In the event of coalescent mastoiditis or cortical erosion of mastoid, cortical mastoidectomy is recommended especially in children after 48 hours of intravenous empirical antibiotics without signs of improvement.⁸ Interestingly, our case showed a sclerotic mastoid bilaterally with soft tissue density within both middle ear which suggest poor aeration and ventilation for a long period, thus the risk of developing complications is higher. Right mastoid cortex was eroded making a direct connection with the temporal area possibly due to the virulence of the bacterium isolated.

Treating Luc's abscess is best managed by surgical approach. Abscess drainage together with myringotomy (with or without grommet insertion) and cortical mastoidectomy was the favored surgery in Luc's abscess.⁶ On the other hand, it is of utmost important to cover suspected Luc's abscess patients empirically with wide spectrum intravenous antibiotics

without waiting for culture and sensitivity tests. Our case was managed with incision and drainage of abscess without myringotomy as the ear canal was edematous which obscured the tympanic membrane. CAT was purposely planned in a different setting to allow complete resolution of temporal swelling before definitive operation addressing the mastoid and middle ear. Decision to perform mastoidectomy is usually guided by pre-operative CT scan. Finally, not doing mastoidectomy or 'mastoid sparing' approach should only be considered if the mastoid was spared, and complete improvement was seen after a simple drainage and antibiotic treatment. Drainage of abscesses is a simple, initial, and conservative approach but less effective compared to mastoidectomy.⁹

CONCLUSION

Luc's abscess is a rare complication of otitis media, and often associated with mastoiditis. Diagnosis is challenging due to the delay signs of suppurative mastoiditis. Thus, HRCT scan should be the investigation of choice to obtain definitive diagnosis and identify associated mastoiditis and rule out possibility of intracranial complications. HRCT findings will help in the decision of performing mastoidectomy in uncertain cases. Cortical mastoidectomy is proven to be more effective than abscess drainage especially in non-responding cases and paediatric group.

CONFLICT OF INTEREST

None to declare.

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