Case Report

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Melioidotic Otomastoiditis in Triplets: A Rare Pediatric Cluster with Intracranial Complications

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Abstract:

Melioidosis, caused by *Burkholderia pseudomallei (B. pseudomallei)*, is endemic in tropical regions, including Thailand. Often called "the great mimicker," it can involve multiple organ systems and present with varied clinical manifestations. While the lungs are the most commonly affected site, head and neck manifestations occur in 14.0–25.0% of cases, primarily as neck abscesses or suppurative parotitis, and otologic involvement is exceedingly rare. We report a cluster of melioidotic otitis media in three 15-month-old triplets, two of whom developed intracranial complications requiring surgical intervention. All patients presented with bilateral otitis media, and melioidosis was confirmed through culture or serologic titer. The two patients with intracranial involvement underwent surgical management and received prolonged treatment with meropenem followed by oral trimethoprim-sulfamethoxazole. At the 7-month follow-up, both surgically treated patients demonstrated complete recovery without chronic sequelae. This case series highlights the importance of considering *B. pseudomallei* infection in refractory otitis media in endemic settings and underscores the need for early recognition of potentially life-threatening intracranial complications.

Keywords: *Burkholderia pseudomallei*, endemic, mastoiditis, melioidosis, otitis media, otologic infections

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Introduction

Melioidosis is caused by the soil- and water-borne, gram-negative organism *Burkholderia pseudomallei* (*B. pseudomallei*)[1]. This formidable opportunistic organism is endemic in tropical and subtropical regions, particularly in Southeast Asia and northern Australia. Immunocompromised individuals, especially those with diabetes mellitus or alcoholism, are at increased risk for melioidosis. Awareness of melioidosis should be heightened during natural disasters, especially flooding, as heavy rainfall and floodwaters facilitate the environmental dissemination of *B. pseudomallei* and increase human exposure, resulting in a surge of cases[2]. Thailand reports over 2,000 culture-confirmed cases of melioidosis annually, representing the highest incidence globally. However, this figure likely underestimates the actual number of human melioidosis cases, which a model study suggests could be as high as 7,572 annually[3]. The mortality rates associated with melioidosis can reach 35.0%[4], largely due to misdiagnosis and under-recognition, particularly in hospitals outside Northeast Thailand, where inadequate training among microbiological personnel may lead to misidentification of the bacteria as a contaminant or another organism[5].

Due to its variable modes and onset of presentation, melioidosis is recognized as "the great mimicker." While bacteremia and respiratory manifestations are more common, head and neck manifestations comprise 14.0–25.0% of cases, predominantly presenting as suppurative parotitis or neck abscesses. In the pediatric populations of Southeast Asia, parotitis is among the most common manifestations of this infection[6], whereas in other regions, such presentations are rare. Otologic involvement is extremely uncommon, with only three cases documented in the literature prior to this report[7-9]. Diagnosis of head and neck melioidosis, particularly otologic manifestations, is therefore challenging due to the rarity of these presentations, non-specific clinical findings, and difficulties in laboratory confirmation, all of which contribute to delayed diagnosis and a potential for serious complications. This report presents cases of melioidotic otomastoiditis in triplets, each exhibiting different degrees of disease severity.

Written informed consent was obtained from the patients' parents for publication of this case series and accompanying images. The institutional review board approved this case series.

Case Report

Case 1

A 15-month-old child, the eldest of triplets, presented to a regional hospital with rhinitis and fever, followed by left-sided otorrhea and dyspnea for 1 week, showing poor response to empirical cefotaxime. The family resided on an island and used groundwater. The patient had no history of recurrent infections or immunocompromised status. Physical examination revealed marked swelling and purulent discharge from the left ear canal, with limited visualization of the tympanic membrane. Computed tomography (CT) of the brain demonstrated bilateral otomastoiditis, a left postauricular abscess, and lateral sinus thrombosis involving the left transverse and sigmoid sinuses (Figure 1). No intracranial abscess was identified.

The patient was diagnosed with complicated bilateral otomastoiditis (including left postauricular abscess and left lateral sinus thrombosis), pneumonia, and respiratory failure requiring endotracheal intubation. Blood cultures yielded *B. pseudomallei*, confirming disseminated melioidosis. Given the severity of complications, the patient was transferred to our tertiary care center for further management.

Upon arrival, the patient underwent urgent left canal wall-up mastoidectomy and bilateral myringotomy with tympanostomy tube insertion. Intraoperative findings revealed extensive purulent discharge and granulation

tissue throughout the left middle ear and mastoid cavity. A Penrose drain was placed in the postauricular region. The patient was successfully extubated on postoperative day 1, and the drain was gradually removed following the cessation of drainage. Microbiological cultures from sputum, left mastoid tissue, and mastoid cavity purulence all isolated *B. pseudomallei*.

Targeted antimicrobial therapy was initiated 1 week after the initial presentation. The regimen consisted of a 6-week intensive phase with intravenous meropenem and co-trimoxazole, followed by a 24-week eradication phase with oral co-trimoxazole. The tympanostomy tube was removed at 2 months postoperatively. Follow-up examinations demonstrated normal bilateral tympanic membranes and age-appropriate hearing thresholds on visual reinforcement audiometry. Anticoagulation with enoxaparin was continued for 3 months for venous sinus thrombosis, with CT venography confirming near-complete resolution. At the 7-month follow-up, the patient demonstrated complete recovery with no residual sequelae.

Case 2

A 15-month-old child, the youngest of the triplets with no prior medical history, presented 1 week after Case 1 with cough and high-grade fever. Following diagnosis of melioidosis in Case 1, meropenem was initiated 2 days after her initial presentation. During hospitalization, she developed right postauricular swelling with erythema and new-onset picket-fence fever, without neurological deficits. Examination showed tympanic membrane bulging, right external auditory canal swelling obscuring the right tympanic membrane, and right mastoid tenderness. Brain CT revealed bilateral otomastoiditis, a right postauricular abscess, right sigmoid sinus thrombosis, and a 0.8-cm rim-enhancing lesion in the right cerebellar hemisphere (Figure 2). Blood cultures confirmed *B. pseudomallei*. The patient was diagnosed with bilateral acute otomastoiditis with postauricular abscess, right lateral sinus thrombosis, and right cerebellar abscess, prompting referral to our tertiary care center for surgical intervention.



Figure 1 Contrast-enhanced brain CT demonstrating a subperiosteal abscess with rim enhancement in the left postauricular region (arrow) and left lateral sinus thrombosis with a filling defect (arrowhead)

Urgent right canal wall-up mastoidectomy with bilateral myringotomy and tympanostomy tube insertion was performed. Similar to Case 1, granulation tissue and purulent discharge were observed in the right mastoid cavity. Following neurosurgical consultation, conservative management was chosen for the subcentimeter cerebellar abscess. Follow-up magnetic resonance imaging (MRI) conducted at 2 days post-surgery confirmed a stable cerebellar abscess, and subsequent MRIs demonstrated complete resolution of the brain abscess 2 months later.

B. pseudomallei was isolated from the right mastoid pus culture, while the mastoid tissue culture yielded no growth. The antibiotic regimen mirrored that of Case 1, consisting of a 6-week intensive phase with meropenem following mastoidectomy, followed by a 24-week eradication phase with oral co-trimoxazole. Enoxaparin was administered twice daily for 3 months until CT venography confirmed the resolution of the thrombosis. Tympanostomy tubes were removed after 3 months. At the 7-month follow-up, the patient remained in good condition with no residual sequelae.

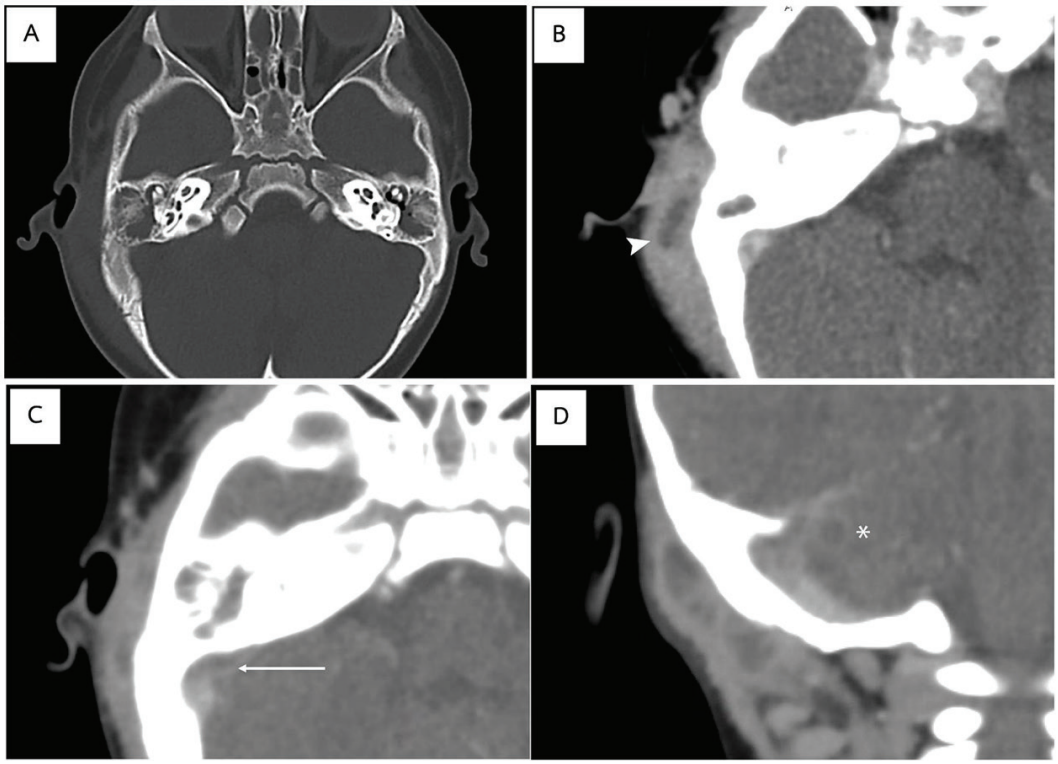


Figure 2 (A) Bone window temporal bone CT showing fluid density in both middle ears and mastoids, with greater severity on the right side (B) Contrast-enhanced brain CT demonstrating a right subperiosteal abscess (arrowhead) (C) Right lateral sinus thrombosis showing the characteristic empty delta sign (arrow) (D) Small right cerebellar abscess (asterisk)

Case 3

A 15-month-old middle triplet infant presented with fever, cough, and rhinorrhea. Contrast-enhanced CT of the brain demonstrated bilateral otomastoiditis without intracranial complications. The patient was diagnosed with uncomplicated bilateral otomastoiditis with pneumonia and sepsis. Hemocultures in this patient were negative; however, a melioidosis serologic titer of 1:80, together with relevant groundwater exposure, temporal clustering, and a clinical presentation similar to the other two cases, supported the high suspicion of melioidosis. Intravenous meropenem was administered during the acute septic phase, followed by ceftazidime. Given the absence of complications requiring tertiary otologic intervention, definitive management was successfully completed at the referring hospital. During the three-month follow-up, the patient was asymptomatic, and the tympanostomy tubes were removed. Supplementary Table 1 summarizes the diagnostic workup across all three cases.

Discussion

Melioidosis is primarily transmitted through percutaneous inoculation, inhalation, or ingestion of *B. pseudomallei* from contaminated soil or water. This transmission route was evident in our cases, where all three triplets were exposed to groundwater at their residences. While melioidosis predominantly affects immunocompromised adults with risk factors, such as diabetes mellitus, chronic kidney disease, and excessive alcohol use, pediatric cases typically occur in immunocompetent children without identifiable risk factors[10].

In Thailand's endemic regions, suppurative parotitis represents the most common presentation of melioidosis in children[6]. This series marks the first reported pediatric melioidotic otomastoiditis, and the first cluster of otologic manifestations, underscoring an urgent need for better awareness of this rare, aggressive presentation. Previous literature describes three cases of melioidotic otomastoiditis, all involving young adults aged 24–35. One example included an immunocompromised patient with diabetes mellitus[9]. The initial case, reported in 1994, involved a Myanmar farmer who presented with left unilateral otorrhea[7]. Pus culture identified *Pseudomonas pseudomallei* (now known as *B. pseudomallei*). The second case documented a Malaysian female with right chronic otitis media complicated by meningoencephalitis and pneumonia[8]. Despite negative pus cultures, diagnosis was established through a significantly elevated indirect hemagglutination assay titer exceeding 1:1,280. The most recent case, reported in 2017, described a 35-year-old male with diabetes who developed severe complications, including mastoid effusion, temporal lobe cerebritis, septic lung metastases, and septic shock[9]. This patient required aggressive surgical intervention with mastoidectomy and craniectomy, followed by extended intravenous antibiotic therapy.

The literature and our experience suggest that melioidotic otomastoiditis represents an aggressive infection with significant risk for intracranial complications. This severity likely stems from delayed diagnosis due to its rarity and non-specific initial presentation, allowing progression to advanced disease. Melioidosis is well recognized for its capacity to progress rapidly from mild or nonspecific symptoms to life-threatening sepsis and death, and this risk appears to be amplified in pediatric patients, who demonstrate substantially higher rates of bacteremia and mortality compared to adults[11]. The triplets in our series demonstrated varying severity of disease, ranging from uncomplicated otitis media to severe complications, including intracranial complications, emphasizing the spectrum of clinical manifestations. Early diagnosis and prompt treatment are crucial to mitigate these complications.

Definitive diagnosis of melioidosis relies on culture isolation from clinical specimens. However, standard culture media have limitations due to *B. pseudomallei*'s

slow growth characteristics and the potential overgrowth by normal flora in non-sterile specimens. Ashdown's selective media enhances detection, allowing identification within 24–48 hours of incubation[12]. Serological testing, particularly indirect hemagglutination (IHA), provides rapid results to complement culture studies. In endemic regions, an IHA cutoff of 1:160 shows 70% sensitivity and 67% specificity, but false positives from prior exposure may limit its utility. In non-endemic settings, a 1:80 cutoff is standard, though lower titers may remain clinically relevant[13]. A study from northeast Thailand has shown that IHA titers increase with age, with children younger than 4 years generally exhibiting lower titers than older children[14]. Therefore, while a titer of 1:80 is not diagnostic, it may be more suggestive of infection in a young child when interpreted alongside compatible clinical and epidemiologic findings. The recent development of PCR assays, both exclusive for *B. pseudomallei* and multiplex assays identifying closely related species, is promising for rapid diagnosis; nonetheless, their inconsistent performance precludes routine use[5]. The CRISPR-Cas12a Test (CRISPR-BP34) is a novel rapid diagnostic tool exhibiting 93.0% sensitivity, delivering results within hours for urine, pus, and sputum specimens, and within one day for blood samples[15]. Our experience with Case 1 illustrates the diagnostic challenge: the initial diagnosis was delayed by one week due to the unusual presentation, while subsequent cases benefited from high clinical suspicion based on contact history.

The organism's intrinsic resistance to penicillins and cephalosporins[1] necessitates specific antimicrobial therapy. Standard treatment comprises an intensive phase with parenteral ceftazidime or meropenem, followed by an eradication phase using oral trimethoprim-sulfamethoxazole to prevent relapse.

Deep-seated infections, such as those observed in Cases 1 and 2, typically require extended intensive therapy[16]. Surgical intervention plays a crucial role in specific situations; while large single abscesses in the liver, muscles, or prostate require drainage, small internal abscesses may respond to medical management alone.[1] In complicated mastoiditis, as demonstrated in our case, mastoidectomy with drainage remains the standard surgical approach. This case series highlights several important aspects: the potential for severe otologic and intracranial complications of melioidotic otomastoiditis, the importance of early recognition and appropriate imaging in suspected cases, and the necessity for combined surgical and medical management in complicated cases. Furthermore, melioidosis should be considered as a differential diagnosis of complicated otomastoiditis in endemic regions, even in immunocompetent children.

Conclusion

Melioidotic otomastoiditis, although rare, represents a significant clinical entity with potentially severe complications if not promptly recognized and treated. While the prognosis is generally favorable with early diagnosis and appropriate management, particularly in localized head and neck infections, the potential for rapid progression and fatal outcomes necessitates high clinical vigilance. This study emphasizes the growing need for physicians, otolaryngologists, and microbiologists to recognize melioidosis as an emerging cause of head and neck infections and abscesses.

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Conflict of interest

The authors declare no conflicts of interest associated with this case series.

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Supplementary Table 1 Summary of microbiologic diagnostic evaluation in the three cases

Diagnostic test	Case 1	Case 2	Case 3
Hemoculture	*B. pseudomallei*	*B. pseudomallei*	Negative
Culture from infection site	1. Left mastoid tissue: *B. pseudomallei* 2. Mastoid cavity pus: *B. pseudomallei*	1. Right mastoid pus: *B. pseudomallei* 2. Right mastoid tissue: No growth	Not reported
Melioidosis titer	Not reported	Not reported	1:80