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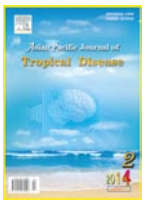

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Bilateral orbital abscesses with subdural empyema and cavernous sinus thrombosis due to melioidosis in a child

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ABSTRACT

Orbital cellulitis is an emergency situation in children. Rapid diagnosis and appropriate management are mandatory to save both vision and life. In contrast, melioidosis infection causing orbital cellulitis with intracranial infection is a rare situation in children, and requires an aggressive management. Poor response or worsening of clinical condition despite appropriate management of paediatric orbital cellulitis should alert the physician of this devastating infection, especially when it occurs in those living in endemic areas.

1. Introduction

Melioidosis is a clinically diverse tropical infection due to the environmental saprophyte *Burkholderia pseudomallei* and is endemic in many parts of South East Asia. The infection usually causes septicemia and results in multifocal infection, especially in those with immunocompromised status. In contrast, localized melioidosis affects healthy individuals and carries a better outcome.

Localized melioidosis occurs as an acute suppurative form, and has been reported to cause parotitis, skin and subcutaneous abscesses, and cervical lymphadenitis in children[1-3]. Eye is not a common site of melioidosis infection in children. A PubMed search revealed only one reported case of melioidosis in a child who presented with a corneal ulcer[4]. The authors describe a case of bilateral orbital abscesses, subdural empyema and cavernous sinus thrombosis due to melioidosis in a child, and the case is successfully treated. Prompt diagnosis, correct choice of antibiotics and early surgical intervention are crucial for good final outcome.

2. Case report

An 11-year-old girl who had no medical illness presented with a history of fever and bilateral periorbital swelling for 3 d prior to admission in a district hospital. Her symptoms were associated with pain and blurring of vision that progressively worsened in that duration. She was treated for bilateral orbital abscesses and was commenced with intravenous cloxacillin 500 mg 6 hourly, cefepime 1 g 12 hourly and metronidazole 500 mg 8 hourly. Computed tomography (CT) scan noted features suggestive of bilateral orbital abscesses (Figure 1A). Despite treatment, her fever persisted and she developed a sudden onset of weakness in her right upper and lower limbs. The patient was subsequently referred to our center for further intervention.

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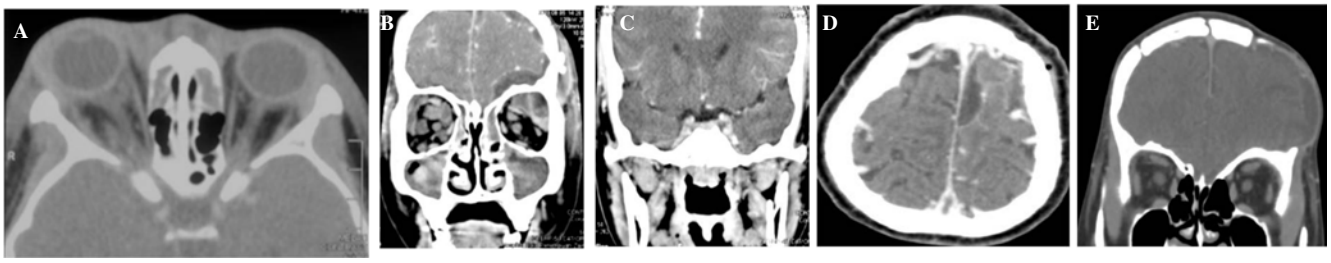

Figure 1. CT scan of the brain and orbit.

A: Initial CT scan of the orbit showed that both globes were proptosed with periorbital soft tissue swelling predominantly on the right side and streakiness of the right retro-orbital fat; B: CT scan of the brain/orbit in coronal images at 1 week later showed rim-enhancing collections in both orbital cavities predominantly on the left side, which is suggestive of orbital abscesses. Intracranial extension was evidenced by subdural empyema in the left frontal convexity; C: A filling defect was observed in the left cavernous sinus with bulging of the lateral wall in keeping with left cavernous sinus thrombosis; D: Hypodense filling defect was observed in the posterior part of the superior sagittal sinus denoting thrombosis and left parasagittal subdural abscess; E: CT scan of the brain/orbit of 2 months later showed that the bilateral orbital abscesses resolved with a small residual non-enhancing subdural collection underneath the left frontal craniectomy site.

Upon arrival at our center, the patient was conscious and alert but lethargic. Her body temperature was 38.5 °C, and no signs of meningeal irritation were observed. She developed a right hemiparesis with an upper extremity power of 3/5 and a lower extremity power of 0/5. The reflexes were brisk, and plantar response was up-going on the right side. Lung and skin examinations were normal.

The patient's visual acuity was 6/36 right eye and 6/60 left eye. There was a 4 mm proptosis bilaterally (Figure 2A). Both conjunctiva were chemotic and congested. A relative afferent pupillary defect was elicited in the left eye. Ocular motility was restricted in all directions of gaze. The cornea was clear, and anterior chamber was deep and quiet. The intraocular pressure ranged between 22-24 mmHg in both eyes. Funduscopic examination was unremarkable bilaterally.

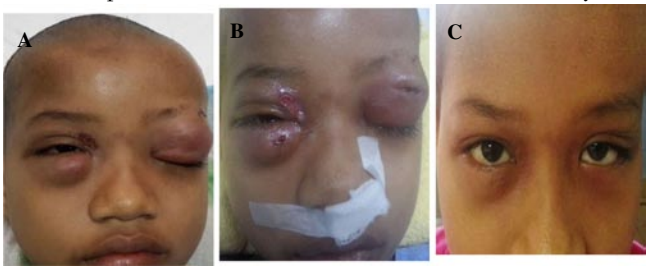

Figure 2. Clinical photographs.

A: Bilateral orbital abscesses during presentation at our center; B: Worsening of orbital abscesses despite intravenous antibiotics; C: Complete resolution of proptosis in both eyes after completing 6 weeks of high dose intravenous ceftazidime.

An urgent repeat CT-scan showed bilateral orbital abscesses with signs of intracranial extension (Figures 1B-1D). The antibiotics were then changed to intravenous meropenem 1.4 g 8 hourly and vancomycin 350 mg 8 hourly. An emergency bifrontal craniotomy was performed to evacuate the pus collections within the subdural space.

Her clinical condition deteriorated a week later. Both eyes became more proptosed (Figure 2B). An incision and drainage of the right lower lid and left upper lid abscesses were performed. The culture from blood and pus did not yield significant growth. The blood picture showed leukocytosis with predominantly neutrophils. Other blood investigations including renal profile, liver function test, blood sugar, coagulation screening and chest X-ray were unremarkable. The abdomen ultrasound did not reveal any evidence of an abscess collection in the liver or spleen.

Diagnosis of orbital melioidosis was considered in view of poor clinical response despite the combination of intravenous antibiotics. Intravenous ceftazidime 525 mg 8 hourly and ceftriazone 1.75 g 12 hourly were commenced with concurrent intravenous meropenem and metronidazole. Intravenous cloxacillin and cefepime were discontinued.

Tuberculosis culture for acid fast bacilli from both the intracranial and orbital specimens was negative. Subsequently, indirect hemagglutination assay (IHA) titer revealed detectable immunoglobulin M for *Burkholderia pseudomallei* (1:160). The patient's antibiotic regimen was then changed to a high dose intravenous ceftazidime 1.2 g 8 hourly. Subsequently, the patient showed a significant clinical improvement. Intravenous ceftriazone, meropenem and metronidazole were then terminated.

The patient's condition gradually improved with progressive resolution of proptosis (Figure 2C). The patient completed the intravenous ceftazidime regimen for a total of 6 weeks duration. A repeat CT-scan of the brain and orbit 2 months post craniotomy showed that orbital abscesses were resolved with a small residual non-enhancing left frontal subdural collection (Figure 1E). The patient was discharged with oral trimethoprim and sulfamethoxazole 960 mg 12 hourly for another 5 months.

At 6 months after completing oral antibiotic regimen, the patient's visual acuity was 6/6 in both eyes and she had full ocular motility in all directions. No sign of relapse or neurological deficit was observed, and the patient ambulated independently.

3. Discussion

Orbital melioidosis with cerebral involvement is an unusual presentation. Four cases of melioidosis causing orbital abscess and intracranial extension have been previously reported in adults[5-8]. Two of these patients died while the remaining two patients survived, but were blind in

the affected eye[5-8]. To date, no similar complications have been reported in paediatric patients. Our patient is an 11-year-old girl who was successfully treated with appropriate antibiotics and surgical intervention, and regained a good final visual outcome with no neurological complications.

Our patient presented with subdural empyema and cavernous sinus thrombosis. Paediatric subdural empyema is a rare, but a life threatening condition. Children commonly present with headache, fever, seizures and focal neurological deficits[9]. The mortality rate ranged from 4.3% to 10.8%[9,10]. A combination of intravenous antibiotics and early aggressive surgical drainage of the pus is known to provide a better final outcome[9,10].

Our patient displayed a poor clinical response during the initial phase despite the combination of antibiotic regimens. The cultures samples were also negative. The IHA titer did not provide an immediate result at our center. Thus, a high index of suspicion is critical when an infection does not appear to respond to conventional antibiotic therapy, especially in the endemic regions.

A cut off value of 1:80 in IHA titer is used to differentiate between true infection and background titer in Malaysia[11]. The diagnostic sensitivity of 1:160 is used in Thailand and Cambodia, while 1:40 titer is of clinical significance in endemic areas of Australia[12-14]. Our clinical diagnosis of melioidosis was presumed based on poor clinical response to conventional antibiotic therapy, borderline IHA titer, and favorable outcome after initiation of ceftazidime regime.

Adult patients with melioidosis infection are reported to have immunocompromised status, which includes diabetes mellitus, renal impairment, retroviral infection, malignancy, steroid therapy and parenteral drug abuse. In contrast, Pongrithsukda *et al.* reported four predisposing conditions for paediatric melioidosis that included dengue hemorrhagic fever, aplastic anemia, thalassemia and G6PD deficiency[15]. Our patient had none of the above predisposing factors except for that she heralds from an endemic population.

Our patient completed intravenous ceftazidime and oral trimethoprim and sulfamethoxazole for a duration of 6 months without serious side effects. Our patient was monitored closely for signs of relapse. Young age, premorbid health status, timely and appropriate antibiotic regimen, and early surgical intervention favor an effective treatment outcome in our patient.

In conclusion, orbital infection with intracranial involvement is an uncommon focus of melioidosis in paediatric patients. Prompt diagnosis and aggressive management are warranted to prevent morbidity and mortality rate in this devastating tropical infection. A high index of suspicion of paediatric melioidosis is necessary as it can be easily missed even in endemic areas.

Conflict of interest statement

The authors declare that we have no conflict of interest.

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References

[1] Stoesser N, Pocock J, Moore CE, Soeng S, Chhat HP, Sar P, et al. Pediatric suppurative parotitis in Cambodia between 2007 and 2011. *Pediatr Infect Dis J* 2012; **31**(8): 865-868.

[2] Lumbiganon P, Chotechuangnirun N, Kosalaraksa P, Teeratakulpisarn J. Localized melioidosis in children in Thailand: treatment and long-term outcome. *J Trop Pediatr* 2011; **57**(3): 185-191.

[3] Pagnarith Y, Kumar V, Thaipadungpanit J, Wuthiekanun V, Amornchai P, Sin L, et al. Emergence of pediatric melioidosis in Siem Reap, Cambodia. *Am J Trop Med Hyg* 2010; **82**(6): 1106-1112.

[4] Siripanthong S, Teerapantuwat S, Prugsanusak W, Suputtamongkol Y, Viriyasithavat P, Chaowagul W, et al. Corneal ulcer caused by *Pseudomonas pseudomallei*: report of three cases. *Rev Infect Dis* 1991; **13**(2): 335-337.

[5] Wong PK, Ng PH. Melioidosis presenting with orbital cellulitis. *Singapore Med J* 1996; **37**(2): 220-221.

[6] Rani A, Fauzi M, Samad Cheung H, Mohd. Shah A, Mahmood T. Melioidosis presenting as orbital and parotic abscess with intracranial extension. *Int Med J Malaysia* 2002; **1**: 1-3.

[7] Jusoh S, Shaharuddin B, Ismail S. Successfully treated rare presentation of orbital melioidosis. *Int J Ophthalmol* 2009; **2**(1): 90-92.

[8] Tirakunwichcha S, Vaivanijkul J. Melioidosis presenting as orbital apex syndrome. *Asian J Ophthalmol* 2009; **11**(1): 40-42.

[9] Legrand M, Roujeau T, Meyer P, Carli P, Orliaguet G, Blanot S. Paediatric intracranial empyema: differences according to age. *Eur J Pediatr* 2009; **168**(10): 1235-1241.

[10] Madhugiri VS, Sastri BV, Srikantha U, Banerjee AD, Somanna S, Devi BI, et al. Focal intradural brain infections in children: an analysis of management and outcome. *Pediatr Neurosurg* 2011; **47**(2): 113-124.

[11] Puthucheary SD. Melioidosis in Malaysia. *Med J Malaysia* 2009; **64**(4): 266-274.

[12] Cheng AC, Peacock SJ, Limmathurotsakul D, Wongsuvan G, Chierakul W, Amornchai P, et al. Prospective evaluation of a rapid immunochromogenic cassette test for the diagnosis of melioidosis in northeast Thailand. *Trans R Soc Trop Med Hyg* 2006; **100**(1): 64-67.

[13] Wuthiekanun V, Pheaktra N, Putchhat H, Sin L, Sen B, Kumar V, et al. *Burkholderia pseudomallei* antibodies in children, Cambodia. *Emerg Infect Dis* 2008; **14**(2): 301-303.

[14] Cheng AC, O'brien M, Freeman K, Lum G, Currie BJ. Indirect hemagglutination assay in patients with melioidosis in northern Australia. *Am J Trop Med Hyg* 2006; **74**(2): 330-334.

[15] Pongrithsukda V, Simakachorn N, Pimda J. Childhood melioidosis in northeastern Thailand. *Southeast Asian J Trop Med Public Health* 1988; **19**(2): 309-316.