CASE REPORT

Systemic Melioidosis Presenting as Suppurative Parotitis

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Received: March 12, 2009;
Review completed: April 15, 2009;
Revision accepted: June 29, 2009.

A 3-year-old child was brought with fever, left parotid swelling and altered sensorium. Cultures from blood, pus and throat swab grew *Burkholderia pseudomallei*. A diagnosis of septicemic melioidosis with encephalopathy was made. She recovered following treatment with parenteral ceftazidime for 14 days, and 6 months of oral co-trimoxazole and amoxycillin- clavulanate. She is doing well on follow-up.

Key words: Abscess, Burkholderia pseudomallei, Melioidosis, Suppurative parotitis.

elioidosis is caused by intracellular bipolar-staining, gram-negative bacillus *Burkholderia pseudomallei*. It was first described in 1911 and is endemic in South East Asia and Northern Australia. Only sporadic cases have been reported from our subcontinent(1,2). Its clinical features mimic that of common infectious diseases like tuberculosis, syphilis and typhoid. We present a child with septicemic melioidosis in view of its rarity.

CASE REPORT

A 3 year old girl was brought with high grade intermittent fever of 8 days duration. On the second day of illness she developed a painful, progressive left parotid swelling. There was also history of purulent discharge from left ear, altered sensorium and abnormal posturing for 2 day prior to presentation. No treatment was sought as the symptoms were attributed to mumps. On arrival, she had an altered sensorium but was able to localize pain, febrile, tachycardic and tachypneic but maintained normal saturation on room air. Her peripheries were cold with feeble pulses and blood pressure was 110/70mmHg, There was no pallor, icterus or generalized lymphadenopathy. She also

had a $7\text{cm} \times 7\text{cm}$ left parotid abscess with purulent aural discharge and an intact tympanic membrane.

Initial investigations revealed polymorphonuclear leukocytosis, hypoalbuminemia and an elevated CRP. The renal parameters, liver function test, CSF analysis, chest skiagram and CT head were normal. A diagnosis of suppurative parotitis, meningoencephalitis with septic shock was entertained. After obtaining two samples for blood cultures, ear swabs and CSF for analysis, the patient was started on ceftriaxone and vancomycin along with fluids, phenytoin, ionotropes and oxygen. On drainage of 30 mL of pus from parotid abscess, it was found to communicate with the left external auditory canal.

On day 3, both the blood cultures reported *Pseudomonas* species and the antibiotics were changed to piperacillin plus tazobactum. This unexpected growth in a community acquired infection, with unusual sensitivity pattern of being resistant to all aminoglycosides and sensitive to amoxyclav, prompted for a careful re-look. Automated bacterial identification system using the mini API-Biomerieux(France), revealed the organism to be *Burkholderia pseudomallei*. The

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antibiotic was then changed to ceftazidime (100mg/kg/day, q8h) on day 4 and continued for 2 weeks. She was afebrile, sensorium improved and the aural discharge decreased, within 48 hours. The cultures from parotid abscess and throat swab also grew *B.pseudomallei*. Screening of the family members with throat swab cultures was negative. At discharge on day 17 she was oriented, ambulant and could speak monosyllables. She received oral eradication therapy with cotrimoxazole (8mg/kg/day of trimetho-prim twice a day) and amoxycillin-clavulanate (40mg/kg/day of amoxicillin thrice a day) for 6 months. Two years post treatment, she is develop-mentally normal with no sequelae.

DISCUSSION

In India, melioidosis is an emerging infection with only few cases reported. A survey near Vellore revealed a seroprevalence of 7%, though serological tests have poor sensitivity and specificity in clinical situations. However, on a population basis, seroprevalence is likely to reflect background exposure to *B. pseudomallei*(3). Cases are also reported amongst European travelers returning from the Indian subcontinent(3). This to a, certain extent ascertains the fact that meliodosis is probably underdiagnosed or under reported due to lack of awareness amongst clinicians and microbiologist.

Burkholderia pseudomallei are a soil and water borne organisms, usually nonpathogenic and have been mainly reported among adults with underlying chronic illness, such as diabetes, thalassemia and chronic renal failure. The infection is acquired through, cutaneous inoculation, inhalation (during heavy rains), ingestion, sexual transmission, amniotic fluid contamination, or can be laboratory acquired(4). The incubation period varies and may remain latent for years. The infection is seasonal in tropics and is common during rains(4). The presentation varies from mild tracheobronchitis to overwhelming cavitary pneumonia, long-standing suppurative focal abscesses, fulminant septicemia, shock and coma, with death being the occasional culminating event. The mortality is over 90% in untreated septicemia, and about 20-75% when treated. Once septic shock develops, the case fatality rate is approximately 95%(5).

In children, localised melioidosis, especially involving the head and neck region is more common than in adults(6). In Thailand, unilateral suppurative parotitis constitutes up to 40% of melioidosis presenting with fever and cheek pain. Most do not have any predisposing factor. Bilateral parotid swelling is rare. The complications reported in partotid swelling are abscess formation, spontaneous rupture into the auditory canal, facial nerve palsy, septicemia and osteomyelitis(3,4,7-9). The overall prognosis for localized infection is good(4,8).

Laboratory identification of *B.pseudomallei* can be difficult, especially in places where it is rarely seen. The large wrinkled colonies in MacConkey agar are usually discarded as contaminants or misidentified as *Chromobacterium violaceum*, *Burkholderia cepacia* or *Pseudomonas aerugi-nosa*. Majority of *B.pseudomallei* isolates are intrinsically resistant to all aminoglycosides, but sensitive to coamoxiclav that helps to differentiate the organism from *P.aeruginosa*(4,6,8,10). In all patients with suspected melioidosis, a throat swab culture should be done as it has a specificity of 100% and a positive culture indicates disease and warrants immediate antibiotic treatment.

The treatment objectives are to reduce mortality and morbidity and to prevent recurrent infection in melioidosis. In children with severe infections, parenteral eradication therapy initial ceftazidime (40mg/kg/dose) or IV meropenem (25mg/kg/dose) eight hourly should be administered for at least 2 weeks. Then a 20 weeks oral maintenance therapy with co-trimoxazole (trimethoprim 8mg/kg/day) and doxycycline (4mg/ kg/day) in two divided doses are recommended. In children below 8 years and preg-nant women, amoxycillin/clavulanate (15mg amoxy-cillin/kg/ dose for three divided dose) is advised instead of doxycycline(4,8). In addition, surgical procedures like aspiration and drainage of pus, curettage and debridement of abscess cavity are required(3,4).

Contact spread is extremely low but reported; hence, cases should be nursed in standard isolation, until they are no longer culture-positive. One week prophylaxis regimen with doxycycline or cotrimoxazole is currently recommended for those SHIVBALAN, et al. Systemic Melioidosis

exposed to heavy contamination (3,4). In our patient, the family, treating physicians and paramedical personnel were given prophylaxis and the child was nursed in isolation. As the infection also has the potential of latency with subsequent reactivation, despite adequate treatment, lifelong follow-up has been advised to the parents.

Contributors: SoS contributed to the concept and design, drafting the article and final approval. He will act as guarantor of the study. VT: collected data, conducted the laboratory tests, and interpreted them. NR & KT analysed the data, helped in manuscript writing and revising it critically.

Funding: None.

Competing interests: None stated.

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