Case report

WHITMORE DISEASE PRESENTING AS PAROTID ABSCESS IN A CHILD

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ABSTRACT

Whitmore disease, alsocalled melioidosis, is an emerging infection in Vietnam after the heavy flood in 2020 with increasing case reports, mostly in adults but also in children. We report a child with parotid abscess, which is one of the common clinical manifestations in the paediatric population in Vietnam. He was previously healthy. The child recovered completely after surgical drainage and antibiotic treatment according to standard guidelines. Because the spectrum of disease in childhood is different from that in adults and underlying predisposing factors are usually not seen in children, a high index of suspicion is required for the diagnosis of paediatric melioidosis. Raising awareness among paediatricians in Vietnam of the clinical presentations of melioidosis in childhood is a prerequisite for improved ascertainment. The key role of the microbiology laboratory in the diagnosis of melioidosis is highlighted.

Keywords: *Melioidosis, Burkholderia pseudomallei, Parotid abscess, Children*

I. INTRODUCTION

Whitmore disease is an emerging tropical disease caused by the Gram-negative bacillus, *Burkholderia pseudomallei*, which resides in soil and surface water [1]. Inhalation of dust contaminated with the organism and percutaneous inoculation or ingestion of contaminated water are considered to be the main modes of transmission [2]. The disease is most common in Southeast Asia and Northern Australia [3]. The endemic area probably extends beyond these two endemic regions as is evident from case reports from Americaand Africa [4]. In Vietnam, an increasing number of cases are being reportedafter the heavy flood in October2020, mostly in adults but also including a few paediatric cases [5]. In this paper, we report a patient admitted toour centerin 2020 with parotid abscesses.

II. CASE REPORT

A10-year-old boy was transferred toour center with a history of fever for 5 days and a painful parotid swelling on the right side of his face for 3 days. The swelling was gradually increasing in size. His past medical history was uneventful and his immunizations were up to date. Cultures were not done at that time and the patient was treated with oral antibiotics. He gave a history of frequent exposure to soil during playing. He lived in the

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area that spent the heavy food in October 2020. He usually bathes from a well and not from any natural water source. There is no history of travel outside.

On examination, he was febrile, with axillary temperatures of 38.5ºC - 39ºC, tachypnoea (respiratory rate of 48/min) and tachycardia (pulse rate of 102/min). Aright-sided tender parotid swelling of 4 cm was present. Other examination findings were normal. Blood investigation results on admission included a C-reactive protein (CRP) of 8.8mg/dl and a white cell count of 7.09 x 10^9/L with 64.6% neutrophils. Ultrasound scan of the parotid swelling showed enlargement of the parotid gland on the right side with heterogenous echogenicity and multiple enlarged intraparotid lymph nodes, the largest being 3.2 cm x 1.2 cm. No abscess formation was noted. The clinical picture was compatible with acute on chronic parotitis with reactive lymphocytosis. The child was started on intravenous cefuroxime and metronidazole for two days but symptoms did not improve and co-amoxyclav was started after omitting the other antibiotics and cefotaxime added after two days.

On day 3, incision and drainage of the parotid swelling was done and pus was sent for culture. On direct Gram stain, no pus cells or organisms were seen. Pus was inoculated on blood, chocolate and MacConkey agar. On the following day, aGram-negative bacillus was isolated on all 3 plates. On blood agar, the colonies were 2-3 mm in size, beta-haemolytic and confluent areas had a metallic sheen. The isolate was oxidase-positive. On MacConkey agar colonies were initially non-lactose fermenting and 1-2 mm, round and convex but later became bright pink. The isolate was considered to be a *Pseudomonas* species and sensitivity testing was done accordingly. The safety pin-like appearance of the isolate further strengthened this possibility. On further sensitivity testing the isolate was found to be sensitive to co-amoxyclav and co-trimoxazole and resistant to colistin, typical of *B. pseudomallei*. Blood from the patient was sent to detect antibodies to *B. pseudomallei.* The isolate was confirmed as *B. pseudomallei* and the child's melioidosis antibody titer by the indirect haemagglutination assay was 160.

The boy was treated with ceftazidime for two weeks in the intensive phase, and was discharged on oral cotrimoxazole and folic acid for 3 months as eradication therapy. His mother was educated on the infection including the importance of continuing oral antibiotics and follow-up to prevent relapse.

III. DISCUSSION

We report a paediatric case of melioidosis presenting with parotid abscess. He were 10 years old. The patient had not any predisposing factors but he had frequent contact with soil, especially, he lived in the area that spent the heavy food in October 2020. He presented with high fever spikes but the leucocyte count and CRP levels were within the normal range.

Melioidosis is more common in adults than in children and infections in children represent only 5-15% of all cases [2,6]. Clinical manifestations also differ between the two groups. Pneumonia is the most frequent presentation in adults and bacteremia is also common. Predisposing risk factors like diabetes mellitus, chronic renal failure, chronic lung and liver disease are commonly (60%-90%) associated with adult patients. In contrast, bacteremia is less common in children, whooften present with skin and soft tissue abscesses, with most having no predisposing factors [7].

Abscess formation, spontaneous rupture into the auditory canal, facial nerve palsy, septicaemia, osteomyelitis and necrotizing fasciitis are reported as complications of melioidosis parotitis [8,9]. Our patient developed a palsy of the marginal mandibular branch of the left facial nerve which improved spontaneously.

The causative organism of melioidosis, *B. pseudomallei*, is a soil saprophyte with the ability

to become a pathogen of both human and animals. It is an aerobic, non-spore forming, motile, Gram-negative bacillus with varying colony morphology. Initially, smooth colonies may become wrinkled with further incubation at 37 °C or room temperature, as shown in one of our isolates.

Concerning laboratory diagnosis, the gold standard is the isolation of the organism from blood or other body fluids of the patient. Although *B. pseudomallei* is not a fastidious organism and grows readily in routine laboratory culture media such as blood agar and Mac Conkey agar, even pure growths are often missed as it is often misidentified. As it is oxidase-positive, it is mostly misreported as *Pseudomonas* species. However, it may sometimes be reported as a coliform due to the bright pink colonies seen on MacConkey agar after 48 hours incubation. A high degree of awareness and familiarity among laboratory staff regarding the colony morphology and the culture characteristics of the organism is therefore needed. Preliminary identification is often made on the grounds of the Gram stain showing the typical 'safety pin' appearance and by demonstrating résistance to gentamicin and other aminoglycosides and colistin or polymyxin B and sensitivity toamoxicillin-clavulanic acid and trimethoprim-sulfamethoxazole. In our first case, we considered the isolate as a *Pseudomonas* species until we noticed the aminoglycoside resistance of the organism.

Confirmation of the identity of the isolate can be difficult. Commercial identification kits such as API 20 NE and Rap Id NF often give incorrect results, identifying the isolate as *Burkholderia cepacia* [10]. The latex agglutination test has been shown to demonstrate high accuracy to confirm the identity of the culture isolate [11]. Confirmation can also be done by PCR [12]. Rapid diagnosis on patients' specimens using PCR and direct immunofluorescence be less sensitive than the culture [1,13]. Antigen detection using a lateral flow immunoassay is a recent development and is expected to give reliable results in pus but not in blood [14].

Presumptive diagnosis using serological tests can be made in both the systemic and localized form of melioidosis as *B. pseudomallei* is capable of producing a humoral immune response in the host. False positives can be seen in the presence of subclinical and asymptomatic infections and also due to cross-reacting antibodies from other organisms [1]. Melioidosis antibody tests were done in our case using the indirect haemagglutination assay and werepositive at slightly elevated titres.

According to the Darwin Melioidosis Guidelines [15], treatment is divided into two phases, the intensive phase and the eradication phase. The drug of choice for the intensive phase is intravenous ceftazidime but for patients in the intensive care unit or with neurological disease, meropenem can be considered. Oral trimethoprim-sulfamethoxazole for 3-5 months is recommended for the eradication phase. Follow-up is important as relapse rates are high if the patient is less compliant during the eradication phase. Some recent case reports of localized melioidosis in children demonstrated successful outcomes without relapses after treating the intensive phase with oral antibiotics [7]. The patient in our study wasregularly followed up every week.

IV. CONCLUSION

Clinical diagnosis of melioidosis in children is often more difficult than in adults because of lack of awareness among pediatricians and because children do not have the common predisposing factors. Therefore, it is important to have a high index of suspicion of melioidosis in any fever of unknown origin or skin and soft tissue infection, especially parotid abscess, as it is the commonest presentation after the heavy flood in October 202. The microbiology laboratory will continue to play a

key role in diagnosing and managing melioidosis.

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