

An Unusual Case Of Sorethroat: Nasopharyngeal Melioidosis

N G Tan, B Ang, D S Sethi

ABSTRACT

A 14-year-old Chinese female presented with severe sorethroat and swinging fever for two weeks despite one course of oral amoxicillin followed by one course of Unasyn (combination of sultamicillin, sulbactam and ampicillin). Throat swab grew *Pseudomonas pseudomallei*. Serology for its antibodies was very strongly positive (>1:512). In this part of the world the IHA titre of 1:16 or greater is significant. She was successfully treated with intravenous ceftazidime. The swinging fever settled within two days. The nasopharyngeal and oropharyngeal lesions cleared after a week of therapy. A further two weeks of Ceftazidime were given to ensure a complete resolution of the infection. Oral tetracycline was given for maintenance therapy. Melioidosis involving various organs have been reported particularly pulmonary melioidosis. Nasopharyngeal melioidosis has not been reported, as far as we know. This is the first reported case.

Keywords: nasopharyngitis, sorethroat, melioidosis

INTRODUCTION

Sore-throat is a common complaint seen by both the family practitioner and the otolaryngologist. The cause of the sore-throat is usually apparent - commonly due to infection either viral or bacterial. Less frequently, the aetiology is non-infective - such as neoplastic, haematological, etc. We report an unusual case of sorethroat - a case of nasopharyngeal melioidosis. This is, we believe, the first reported case of melioidosis of the nasopharynx in the English language medical literature. This conclusion was based on a search of the medical literature published over the last twenty years.

Melioidosis is the name given to all diseases caused by the bacterium *Pseudomonas pseudomallei* (also called *Burkholderia pseudomallei*). The organism can infect any organ, although the lung is the most common organ affected⁽¹⁾. The first case of Melioidosis of the parapharyngeal space has been reported in 1991⁽²⁾.

CASE REPORT

A 14-year-old Chinese female student presented with a two-week history of severe sorethroat, high swinging fever and postnasal drip. There were also associated symptoms of neckache and stiffness,

earache and occipital headache. She has a positive history of exposure to soil at a local beach 3 weeks prior to her symptoms. She was treated by her family practitioner as a case of acute pharyngotonsillitis. She was given two courses of antibiotics (amoxicillin followed by Unasyn, combination of sultamicillin, sulbactam and ampicillin) over a two-week period. Her condition did not improve and she was subsequently referred by her family practitioner to our department.

On clinical examination, she was toxic with high swinging fever. Nasopharyngoscopy showed that she had severe infection - her adenoids were enlarged, grossly inflamed and was covered with a coat of mucopus (Figs 1 and 2). The infection had involved the oropharyngeal region where the tonsils and posterior pharyngeal walls appeared granulomatous and swollen, and were also covered with purulent exudate. She also had bilateral cervical lymphadenitis with tender lymph nodes with sizes ranging from 1 to 2 cm. Endoscopic examination of the nasal cavity was done to ensure an excellent view of both the anterior and posterior nasal spaces. In this patient, it was done also because she could not tolerate the postnasal space examination with the routine PNS mirror.

A swab culture of throat organisms grew the caustic organism - *Pseudomonas pseudomallei*. Serology for its antibodies was very strongly positive (> 1: 512 titre). In this part of the world the IHA titre of 1:16 or greater is significant⁽³⁾.

She was successfully treated with a three-week course of high dose intravenous ceftazidime. She responded promptly as shown by the abolition of her high swinging febrile state within two days of starting ceftazidime. The nasopharyngeal and oropharyngeal lesions cleared after a week of therapy and a further two weeks of ceftazidime were given to ensure a complete resolution of her melioidosis infection. She was given oral tetracycline for maintenance therapy after her discharge from hospital and has been regular in appointments for surveillance for relapse.

DISCUSSION

History of Melioidosis

Melioidosis is a glanders-like disease in humans and animals known for the past 80 over years since it was first described by Whitmore and Krishnaswami⁽⁴⁾. It was then called the 'Whitmore disease'. The name

Department of Otolaryngology
Singapore General Hospital
Outram Road
Singapore 169608

N G Tan, MBBS, FRCS, DLO
Consultant

D S Sethi, MBBS, FRCS, FAMS
Consultant

Department of
Infectious Diseases
Communicable Disease Centre
Tan Tock Seng Hospital
Moulmein Road
Singapore 308433

B Ang, MBBS, FRCP, FAMS
Consultant

Correspondence to:
Dr N G Tan

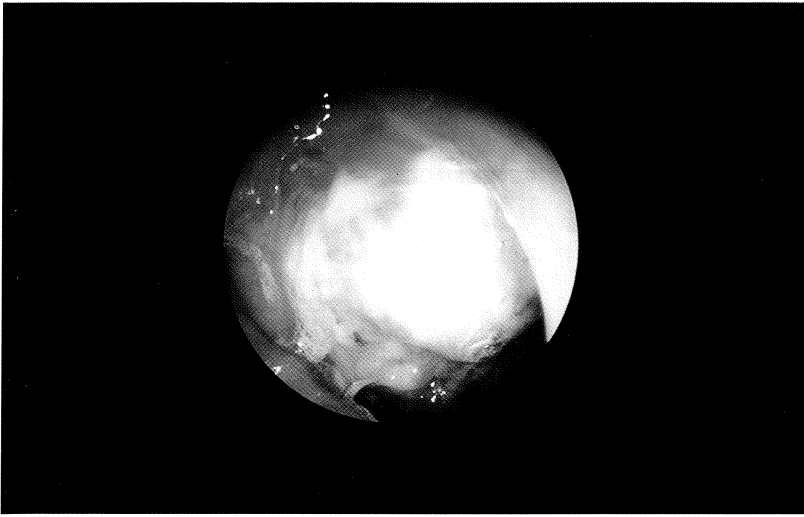


Fig 1 - Endoscopic view of the nasopharynx through the left nasal cavity by a zero Storz degree endoscope. The adenoid pad is grossly enlarged and inflamed and is covered with a layer of mucopus. The infection extended into the oropharyngeal region.



Fig 2 - Endoscopic view of the nasopharynx after 3 weeks of intravenous ceftazime. The mucopus covering the adenoid pad has resolved and the pad has shrunken in size to that consistent with her age.

for this disease Melioidosis ('condition resembling the distemper of asses') however was first coined by Stanton and Fletcher⁽⁵⁾ in 1921. The causative organism has been called *Pseudomonas pseudomallei* and it had many names previously – *Bacillus pseudomallei*, *Pfeifferella whitmori*, *Bacillus whitmori*, *Flavobacterium pseudomallei*, *Actinobacillus pseudomallei*, *Loefflerella* and *Malleomyces pseudomallei*. It is recently proposed that this organism be put into a new genus *Burkholderia*⁽⁶⁾.

Epidermiology of melioidosis

The disease is prevalent in lands within the latitudes of 20°N and 20°S of the equator⁽⁷⁾. It is endemic in Southeast Asia, particularly Thailand, northern Australia, India, Hong Kong, and Taiwan⁽⁸⁾. Reports of sporadic cases in temperate countries such as Japan, USA, UK etc, revealed that the patients usually have travelled to or immigrated from the endemic countries⁽⁹⁾.

B. pseudomallei is a free-living, Gram-negative, obligatory aerobic, non-sporing forming soil bacillus found most frequently in surface waters and during the raining seasons⁽¹⁰⁾. It is now known that the mode of transmission is by exposure to the free-living *B. pseudomallei* organism in the environment. The two common ways for this to happen: by direct contact with contaminated soil and water through skin cuts and abrasions and inhalation of dust particles containing the bacteria⁽¹¹⁾.

It is difficult to determine the exact length of the incubation period. Though many cases are reported with three weeks of exposure to soil or surface waters, there were reports in non-endemic countries of ex-soldiers or travellers manifesting the disease after 10 to 20 years later⁽¹²⁾. Certain predisposing factors are noted, these include diabetes mellitus, chronic renal failure, malignancy and of course, occupational exposure⁽¹³⁾.

Clinical presentation

The case reported in this paper is an example of a localised infection in the form of an acute suppuration. Parapharyngeal melioidosis is another such example. There is another way that localised infection can present itself and that is the chronic granulomatous lesion eg a chronic skin granuloma. Apart from local infections, melioidosis can present in another two ways. One way is as a septicaemia of abrupt onset. With dissemination of the primary focus of infection frequently evidenced clinically by the development of multiple subcutaneous abscess, pulmonary shadows, joint swelling and myositis. The other way, as a prolonged fever, with or without a demonstrable infectious site⁽¹⁰⁾.

It should be noted however that symptomatic melioidosis is in the minority of patients. The majority of infected patients are asymptomatic. Illness may be delayed for years because of the unique ability of *P. pseudomallei* to remain quiescent in infected individuals. The factors that influence reactivation of the dormant pathogen are not well understood but probably include environmental variables, stress and immunity status.

Diagnosis of melioidosis

The clinical presentations of melioidosis can be so varied and unusual and the need for rapidity of an accurate diagnosis is required to ensure the survival of the patient. Melioidosis has been termed "the great imitator" of every infectious disease, as virtually any organ can be affected. Its chronic form has been mistaken for anaerobic infections, tuberculous or fungal infections, and the pyogenic form of *Staph. aureus* of other acute bacterial infections. Histopathologic study has shown the formation of abscess in the acute stage and granuloma in the chronic form. Definitive diagnosis can be made from cultures of secretions or pus from the sites of

infection. The organism grown on the bacteriological media is then isolated and undergo biochemical tests (with commercial test strips like the API20NE and Microbact⁽¹⁴⁾) to confirm the identity.

Serological test is used as an adjunct, as there are limitations to its interpretation which depends on the seroprevalence, especially in endemic areas⁽¹⁵⁾. Three serological methods have been found to be reliable:

1. Indirect haemagglutination (IHA) test
2. Immunofluorescence detection of specific IgM antibodies
3. IgM-ELISA test

Other serological tests:

4. The gold blot detection of IgM and IgG-specific antibodies
5. Immunosorbent assay of *B. pseudomallei* exotoxin
6. Avidin-biotin enzyme-linked immunosorbent assay of *B. pseudomallei* antigens

Treatment of melioidosis

Management of melioidosis consists predominantly, of early, appropriate and adequate antibiotics regimes. Those antibiotics which are moderately active include chloramphenicol, co-trimoxazole, kanamycin, novobiocin, tetracycline, tetracycline congeners (eg doxycycline, rolitetracycline, minocycline) and penicillin with B lactamase inhibitors⁽¹⁶⁾. The recommended antibiotics for the more severe cases include ceftazidime, imipenem-cilastin, piperacillin, azlocin and carumonam. Combination therapy is advisable to minimise the problem of antibiotic resistance⁽¹⁷⁾. After the resolution of the acute infection, the patient is usually put on maintenance therapy for three to six months.

CONCLUSION

Melioidosis with its protean manifestations and propensity for any organ system is potentially fatal and difficult to diagnose. Clinicians must have a high index of suspicion in cases of unexplained prolonged febrile illness and always consider the possibility of this disease. This case report illustrates an insidious and unusual presentation of melioidosis – nasopharyngeal melioidosis.

ACKNOWLEDGEMENT

Our thanks to Dr Cecilia Ngan and staff of Diagnostic Bacteriology Laboratory, SGH

REFERENCES

1. Ip M, Osterberg CG, Chan PY, Raffin TA. Chest 1995; 108(5): 1402-4.
2. Elangos, Sivakumaran S. Parapharyngeal space melioidosis in a diabetic. J Laryngol Otol 1991; 105: 582-3.
3. Yap EH, Chan YC, Ti TY, Thong TW, Tan AL, Yeo M, et al. Serodiagnosis of melioidosis in Singapore by the indirect haemagglutination test. Singapore Med J 1991; 32:211-3.
4. Whitmore A, Krishnawami CS. An account of the discovery of a hitherto undescribed infective disease occurring among the population of Rangoon. Indian Med Gaz 1912: 47:262-7.
5. Stanton AT, Fletcher W. Melioidosis, a new disease of the tropics. Trans 4th Congress Far Eastern Assn Trop Med 1921; 2:196-8.
6. Yabunche E, Kosako Y, Oyaizu H, Yano I, Hotta H, Hashimoto Y, et al. Proposal of Burkholderia gen. Nov. and transfer of seven species of the genus Pseudomonas homology group II to the new genus, with the type species Burkholderia cepacia comb. Microbiol Immunol 1992; 36:1251-75.
7. Howe C, Sampath A, Spotnitz M. The pseudomallei group: a review. J Infect Dis 1971; 124:598-606.
8. Lee N, Wu JL, Lee CH, Tsai WC. Pseudomonas pseudomallei infection from drowning: the first reported case in Taiwan. J Clin Microbiol 1985; 22:352-4.
9. Spotnitz M, Rudnitzky J, Rambaud JJ. Melioidosis pneumonitis: analysis of nine cases of a benign form of melioidosis. JAMA 1967; 202:950-4.
10. Leelarasamee A, Bovomkitti S. Melioidosis: review and update. Rev Infect Dis 1989; 11:413-25.
11. Thin RNT, Brown M, Stewart JB, Garrett J. Melioidosis: A report of 10 cases. Quart J Med 1970; 39:115-27.
12. Mays EE, Ricketts EA. Melioidosis: recrudescence associated with bronchogenic carcinoma twenty-six years following initial geographic exposure. Chest 1975; 68:261-3.
13. Guard RW, Khafagi FA, Bridgen MC, Ashdown LR. Melioidosis in far north Queensland. A clinical and epidermiological review of twenty cases. Am J Trop Med Hyg 1984; 33:467-73.
14. Dance DAB, Wuthiekanum V, Naigowit P, White NJ. Identification of Pseudomonas pseudomallei in clinical practice: use of simple screening tests and API 20NE. J Clin Pathol 1989; 42:645-8.
15. Embi N, Suhaimi A, Mohamed R, Ismail G. Prevalence of antibodies to Pseudomonas pseudomallei exotoxin and whole cell antigens in military personnel in Sabah and Sarawak, Malaysia. Microbiol Immunol 1992; 36:899-904.
16. Ashdown LR. Evaluation of antibiotics in Pseudomonas infections. Med J Aust 1979; 1:284.
17. Sookpranoee T, Sookprane M, Mellencamp MA, Prehwim LC. Pseudomonas pseudomallei, a common pathogen in Thailand that is resistant to the bactericidal effects of many antibiotics. Antimicrob Agents Chemother 1991; 35:484-9.