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CONCISE ARTICLE

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A Case of Tuberculous Sinusitis Without Concomitant Pulmonary Disease

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Abstract This case report highlights the difficulty of diagnosing tuberculous sinusitis in the absence of pulmonary foci. Although extrapulmonary localisations of tuberculosis are rare in immunocompetent patients, it is important to consider this diagnosis, since therapeutic delay usually results in an unfavourable outcome. Acid-fast bacilli are sometimes difficult to detect in pathological specimens. Consequently, the diagnosis is usually based on the following criteria: (i) the absence of clinical response to usual antibiotics, (ii) the presence of caseous granulomatous inflammatory lesion on histopathology, and (iii) identification of *Mycobacterium tuberculosis* by the polymerase chain reaction assay confirmed by bacteriological culture. The diagnosis of tuberculosis is finally confirmed by the efficacy of antituberculous treatment. The differential diagnosis is Wegener's disease.

Introduction

The tubercle bacillus was discovered by Koch in 1882, and one of the first reviews of upper respiratory tract tuberculosis and its treatment with streptomycin was published in 1951 [1]. Until 1985, the number of cases of pulmonary tuberculosis in the USA decreased every year, whereas the number of extrapulmonary cases remained stable. The percentage of cases due to extrapulmonary disease increased along with the rise of human immunodeficiency virus (HIV) coinfection, and in 1991 21% of extrapulmonary cases in the USA were associated with HIV infection. However, ear, nose and throat localisations are rare and unrecognised [2]. Here, we report a case of tuberculous sinusitis without concomitant pulmonary disease in a non-HIV-infected patient.

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Case Report

In December 1999, a 49-year-old Rwandan woman was admitted to our hospital with sinusitis unresponsive to standard therapy. She had no history of past disease or allergy and was not taking any medication. She had received the BCG vaccine 10 years previously. She was neither a smoker nor an alcoholic. Her niece had suffered from pulmonary tuberculosis 3 years earlier and was treated successfully.

The patient's symptoms of nasal congestion with facial pain and fever began 3 months prior to admission. A physician had established a diagnosis of sinusitis and prescribed oral ofloxacin and intramuscular ceftriaxone for 6 days. Due to lack of improvement, this treatment was discontinued and switched to oral pristinamycin and corticotherapy (20 mg daily). Two weeks later, the symptoms persisted and a computed tomography scan was performed, revealing bilateral maxillary and right ethmoid sinusitis. The frontal and sphenoidal sinuses had normal ventilation. Because nasal obstruction was persistent in spite of empirical treatment, biological, histological, and bacteriological diagnostic procedures were performed.

The results of routine laboratory tests, including haemoglobin levels, leucocyte and platelet counts, glucose, electrolyte, creatinine, blood urea, and serum protein levels, were all normal. The patient's C-reactive protein level was 32 mg/l. Culture of the nasal discharge was screened for atypical infectious agents and resistant strains of bacteria, but microscopic examination showed no neutrophils or bacteria. Since this diagnostic approach was unrevealing, further evaluation was necessary.

Fiberoptic nasoscopy was performed with histological, bacteriological and mycological sampling. Histologic examination revealed several granulomata with necrosing vasculitis compatible with Wegener's disease or tuberculosis. An etiological test for antineutrophil cytoplasmic antibodies was negative. Direct examination on Ziehl-Neelsen stain revealed numerous acid-fast bacilli. Polymerase chain reaction identified *Mycobacterium tuberculosis*. Bacteriological culture confirmed this diagnosis, and the patient's tuberculin reaction was positive.

In order to identify all potential foci of tuberculosis, further examinations were performed with the following results: aspiration of gastric contents and urine cultures were negative, chest radiographs and computed tomography scans were normal. Since no other foci of tuberculosis were found, we suspected the patient to have an immune deficiency, and further tests were performed with the following results: HIV test was negative, CD4+ and CD8+ cell counts were normal (612/mm^3 and 178/mm^3, respectively), immunoelectrophoresis of serum proteins was normal, and Bence jones' protein was not detected in urine. Therefore, the diagnosis of tuberculous sinusitis with no other foci or underlying disease was established.

Susceptibility testing performed on the isolated organism revealed sensitivity to isoniazid and rifampin. The patient was consequently treated with a 9-month course of antituberculosis therapy, i.e., a 4-month course of isoniazid, pyrazinamide, rifampin, and ethambutol and a 5-month course of rifampin and isoniazid. Eighteen months after the end of antituberculous treatment the patient was doing well and bacteriological samples were negative.

Discussion

Extrapulmonary tuberculosis currently accounts for 25% of tuberculosis cases in France. Although all organs are involved, the most frequent extrapulmonary localisations are the lymph nodes [3]. Tuberculosis of the upper respiratory tract is infrequent [4], especially extralaryngeal forms including oropharyngeal, otitic and sinusal localisations [5, 6]. The sinusal anatomy and the aerial mode of spread of the bacillus should lead us to think that tuberculous sinusitis is not so rare and might in fact be underdiagnosed, either in association with or without a pulmonary localisation.

Diagnosis of extrapulmonary tuberculosis is often difficult, except in cases of concomitant pulmonary involvement, and surgery is usually required for biopsy [5, 7]. Diagnosis of sinusitis tuberculosis is even more difficult because acid-fast bacilli are difficult to detect in pathological specimens, and the diagnosis is usually based on the efficacy of specific antituberculous medications [8]. It is therefore important to think about this unusual diagnosis in cases of sinusitis that do not respond to empirical therapy.

In the case presented here, the course of the patient's illness was not typical of chronic bacterial sinusitis. First, the microbiological investigation did not reveal a common bacteria. Second, the patient's response to the antibiotics typically prescribed for this condition was unsatisfactory. Finally, the patient's country of origin and the history of tuberculosis in the patient's family suggested a tuberculous infection. A tropical parasitologic or mycologic infection would also have been possible.

In our case, the diagnosis of tuberculous sinusitis was made by histological examination, which is often indispensable for establishing the diagnosis of such an atypical localisation [9]. However, attention must be drawn to the fact that in cases of tuberculous sinusitis without a concomitant pulmonary localisation, the differentiation between the histological diagnosis of tuberculosis versus Wegener's disease is complex and often faulty [10]. The antineutrophil cytoplasmic antibody test helps establish the diagnosis, although the result is negative in 15% of localised Wegener's granulomatoses.

Another diagnostic difficulty exists with regard to HIV-infected patients. In these patients the diagnosis of tuberculosis is complicated by their decreased tuberculin reactivity and atypical chest radiography [11]. Nevertheless, the incidence of extrapulmonary tuberculosis doesn't seem to be attributable to HIV infection [12]. Previous studies have shown that disease progression is more rapid in these patients, but the response to therapy mirrors that of control patients [11].

The present report adds to the literature a new case of a rare extrapulmonary localisation of tuberculous that occurred in an immunocompetent patient. Sinusal tuberculosis is difficult to diagnose but must be considered when a case of sinusitis resists common treatment, especially in persons with a history of contact with a tuberculosis-infected person. Appropriate imaging studies and examination of bacteriological and histological samples are key to making a correct diagnosis. Awareness of this disease entity is very important since therapeutic delay often has a negative outcome. Response to antituberculous medical treatment is usually satisfactory.

References

1. Omerod et al. (1951) A review of tuberculosis of the upper air passages during the past thirty years and its treatment by streptomycin. J Laryngol Otol 65:461–471
2. David W, Haas R, Des Prez M (1995)Mycobacterium tuberculosis. In: Mandell GL, Bennett JE, Dolin R (eds) Principles and practice of infectious diseases. Churchill Livingstone, Philadelphia, PA, pp 2213–2240
3. Stelianides S, Beltamoug N, Fantin B (1997) Manifestations and diagnosis of extrapulmonary tuberculosis. Rev Mal Respir 14 (Suppl 5):72–87
4. Lau SK, Kwan S, Lee J, Wei WI (1991) Source of tubercule bacilli in cervical lymph nodes: a prospective study. J Laryngol Otol 105:558–561
5. Fortun J et al. (1996) Tuberculosis of the otorhinolaryngologic region: laryngeal and extra-laryngeal forms. Enferm Infecc Microbiol Clin 14:352–356
6. Scully RE, Mark EJ, Mc Neely WF, Ebeling SH (1999) Case records of the Massachusetts general hospital. N Engl J Med 340:1349–1354
7. Klaudel A, Gawlicka G, Spyra W, Wiewiora A (1992) A rare case of tuberculosis of the maxilla, zygomatic region, maxillary sinus and facial skin in a patient with diabetes mellitus. Wiad Lek 45:317–319
8. Khalil M, Lindley S, Matouk E (1985) Tuberculosis of the orbit. Ophthalmology 92:1624–1627
9. Swart JG, Flamingh DQ de, Hamersma T (1987) Histologically detected extrapulmonary tuberculosis in the head and neck region. A review of 222 cases. S Afr Med J 11:700–702
10. Windler B (1989) A rare case of an extra-pulmonary tuberculosis. Offentl Gesundheitswes 51:505–509
11. Shafer RW et al. (1991) Extrapulmonary tuberculosis in patients with human immunodeficiency virus infection. Medicine 70:384–397
12. Cowie RL, Sharpe JW (1997) Extra pulmonary tuberculosis: a high frequency in the absence of HIV infection. Int J Tuberc Lung Dis 1:159-162