Naso-Sinusal Tuberculosis: 3 Cases Report

Ali Mardassi 1,*, Hela Dimassi 1, Safa Nefzaoui 1, Sonia Benzarti 1

1 ENT Department, Military Hospital of Tunis, Tunisia

* Corresponding author: Ali Mardassi, Assistant Professor, ENT Department, Military Hospital of Tunis, Tunisia. E-mail: alimardassi@gmail.com

Abstract

Introduction: Tuberculosis remains a real public health problem in Tunisia. Extra-pulmonary localizations account for 20 to 30% of tuberculosis disease, dominated by the node, osseous and urogenital localization.

Case Report: The authors present three cases of nasosinusal tuberculosis, an unusual and rare localization of the disease, in a man and 2 women aged respectively of 30, 65 and 36 years. Results: The main symptoms were dominated by nasal obstruction, posterior throwing and dental pain. CT-scan showed a filling of the nasosinusal cavities with various stages of bony lysis. In all the cases, tuberculosis was confirmed histologically after an endoscopic middle meatotomy. Investigations looking for tuberculosis elsewhere were negative in all cases. Treatment was based on anti-tuberculosis drugs during 9 to 12 months. The evolution was favourable in all the cases.

Conclusions: Nasosinusal tuberculosis is a rare chronic infection associating often variable levels of bony lysis. Histological exam remains the best way to confirm it. Therapy is based on anti-tuberculosis drugs. Medical investigations must usually search other localizations of the disease.

INTRODUCTION

Tuberculosis (TB) remains a major public health problem worldwide, with 9 million new cases reported the World Health Organization (WHO) in 2013. The mortality was about 1.5 million [1]. In Tunisia, it remains a frequent disease affecting especially poor classes. Extra pulmonary localizations account for 20 to 30% of Tuberculosis disease, dominated by the lymph nodes, osseous and urogenital sites [2]. It unusually involves the nose and the paranasal sinuses which are rarely the primary site of the infection [3, 4].

CASE REPORT

We report three cases of extra-pulmonary tuberculosis involving the maxillary sinuses. All the patients were followed and treated at the ENT department of the Military Hospital, Tunis, Tunisia.

Case 1

A 36 year old lady presented with nine months history of right nasal blockage associated with a purulent discharge. Nasal endoscopy showed a granulomatous, congestive and bleeding mucosa fulfilling the entire right nasal cavity. The same findings were noted at the nasopharynx. Laboratory tests found a sedimentation rate of 75. The white blood cells count was of 5800. We noted a positive tuberculin skin test result (10 mm induration). CT scan of paranasal sinuses showed a total filling of the right maxillary and ethmoid sinuses (Fig 1).

Figure 1: CT scan of the paranasal sinuses in coronal bone window section, showing a total filling of the right maxillary sinus (yellow arrow) associated to a roof hyperplasia of the ethmoidal sinus (red arrow)

A right middle meatotomy was performed, associated to a nasal, sinusal and nasopharyngeal mucosa biopsy. Histological examination found caseating granulomatous lesion confirming the diagnosis of a nasopharyngeal and rhino-sinusal location of tuberculosis.
Investigations looking for other sites of Tuberculosis (Sputum bacilloscopy, bronchoscopy, chest X-ray and urine culture) were negative. She was treated with antituberculosis drugs. The therapeutic protocol consisted on a quadruple therapy for 2 months; Isoniazid (5 mg/kg); Rifadin (10 mg/kg), pyrazinamid (25 mg/kg) and (streptomycin 1 g/day), followed by a double therapy (Isoniazid and Rifadin) during 8 months. Regular follow up has noted a progressive regression of the initial physical complaints. Endoscopic control showed a thickened mucosa. A nasosinusal biopsy was performed at the end of treatment and hasn’t shown any specific pathological lesions. Endoscopic and radiological controls after 3 years of follow up were also favorable and didn’t note a recurrence of the disease.

Case 2
A 65-year-old diabetic woman treated for hypertension, complaining of left suborbital pain and nasal obstruction. These symptoms were associated to anosmia without rhinorrhea or epistaxis history. A weight loss of 10 Kilograms over a period of two months was reported. The clinical presentation occurred four months after a tooth extraction. Endoscopy showed a unilateral suspicious polypoid lesion at the level of the left middle meatus. Physical examination did not find cervical lymph nodes. Laboratory tests revealed an accelerated sedimentation rate and a high white blood cell count (12500). Tuberculin skin test was negative. CT scan of the paranasal sinuses revealed a total filling of the left maxillary sinus, bone lysis of the anterior and the medial walls of the maxillary sinus associated to an erosion of the malar bone (Fig 2).

A biopsy of the sinonasal formation through a middle meatotomy was performed. Histopathologic examination concluded to specific inflammatory lesions of tuberculosis. Investigations searching for tuberculosis elsewhere were negative. The patient underwent a quadruple antituberculosis therapy (Isoniazid, Rifadin, Pirazinamid, Ethambutol) for two months, followed by a double therapy (Isoniazid, Rifadin) for 7 months. The regular follow-up noted a regression of the clinical symptoms, a satisfactory endoscopic findings and a normalization of the biological tests. A one-year CT scan control showed a vacant sinus cavity with bony regenerations sites (Fig 3).

Case 3
A 30-year-old man, heavy smoker, presented with a four months history of maxillary and dental pain resistant to medical treatment. Physical examination noted a poor oral health and a loss of the palate mucosa exposing the bone at the level of the 24 and the 25 teeth which were mobile. Endoscopy found a granulomatous nasal mucosa with a free middle meatus. CT scan of paranasal sinuses objectified a partial filling of the left maxillary sinus associated to an erosion of its floor and a bony sequestrum opposite to 24, 25 and 26 teeth (Fig 4).

Laboratory tests were normal except for a moderate lymphocytosis. The tuberculin skin test result was positive (10 mm induration). A dental extraction of the 24 and 25 teeth was performed, and was associated to the removal of the sequestrum and a biopsy of the left maxillary sinus (Fig 5). Histological examination revealed foci of tuberculoid lesions with caseous necrosis. Basic investigations searching for other TB sites (Sputum bacilloscopy, bronchoscopy, chest X-ray and urine culture) were negative. Treatment was based on a quadruple therapy during 2 months followed by a double antituberculosis chemotherapy for 10 months. The follow up showed a regression of granulomatous lesions of the mucosa and a decline of the dental and jaw pain. An improvement of the general symptoms was also noted with asthenia disappearance and weight gain. The endoscopic control was normal after a follow-up of 1 year. The oro-antral communication was repaired using a fibro-mucosal flap associated to the Bichat fat.
DISCUSSION

Tuberculosis (TB) remains a major global health problem affecting a third of the world's population [1]. After a sharp decline of Tuberculosis until 1985, its incidence is rising in the 1980s with the emergence of HIV, increasing population, poverty and migration of populations [5, 6].

Brazil, the Russian Federation, India, China and South Africa (BRICS), collectively account for almost 50% of global TB cases [1]. Literatures review showed that tuberculosis involving nose and paranasal sinuses is extremely rare and usually arises from tuberculosis elsewhere, like in the respiratory tract [5-7]. However, it is necessary to evoke it in the presence of any infection or chronic inflammation of nasosinusal cavities or if facing diagnostic difficulties [8-11].

The primitive contamination of the nasosinusal cavities is still rare given the strict aerobic nature of Koch bacillus [8].

All sinuses can be attacked by tuberculosis, but the maxillary sinus and the ethmoid cells are the most affected [6]. According to other authors, the sphenoid localization occurs mainly in children, although it is rare [12]. In our study, the infection was localized in the maxillary sinus in all patients.

The assumed mode of contamination of these regions is multiple [13]:
- Local inoculation by inhalation of infected particles (nasopharynx, nasal cavity, nasopharynx). Inoculation can occur from the outside environment, but also from an endogenous pulmonary focus.
- Local traumatic digital inoculation (nasal localization).
- Inoculation through blood or lymphatic system from an unknown site (false appearance of a primitive localization), or from a recognized site (pulmonary for the hematogenous route, mucosal for the lymphatic one).

Concerning histopathology, there are essentially three types of lesions [14, 15]:
- The larval form where there is a mucosal involvement causing a mild sinusitis. The antrum is filled with polyps and thickened mucus with a swampy pale appearance. Purulent secretions are scarce and the diagnosis is difficult. This is the most common macroscopic presentation.
- Chronic sinusitis with bone involvement. Purulent secretions are abundant and Mycobacterium tuberculosis (BK) is easily found.
- The hyperplastic type is responsible of the formation of tuberculoma.

Naso-sinusal localization is unusual and is rarely the primary site of TB infection [8, 9]. In our patients, there was a history of smoking and uncontrolled diabetes. Tobacco and immunosuppression factors are thought to enhance the prevalence of tuberculosis [16]. The presenting complaints are nonspecific dominated by nasal obstruction, rhinorrhea and jaw pain, suggesting a banal chronic sinusitis [5]. Anosmia is rare [3] and was noted in one of our patients. However, unilateral lesions, non response to medical treatment, long-term evolution and the possible association with a decline in general health should alarm [17].

Similarly, there are no specific endoscopic aspects of TB infection. Budding lesions, mucosal or granulomatous hyperplasia have been described [3, 13]. In our study, the endoscopic look of the middle meatus lesion was suspicious in one case. Histological examination after a biopsy confirms the diagnosis of tuberculosis.

Radiological assessment mainly involves a CT scan of the facial bones with axial and coronal sections. Radiological elements are not specific too; the main described finding is a filled sinus, associated in some cases, to a parietal bone lysis. The interest of the imaging is mostly to establish a lesional balance sheet and a control of the processing under treatment [6, 9, 18].

The biopsy of the maxillary sinus followed by histological examination is considered to be the most reliable proof of the diagnosis [4]. This biopsy can be done either by endonasal or endobuccal approach [19]. Clement noted that tuberculosis should be suspected in presence of chronic inflammation granuloma with epithelial cells and lymphocytes, even without caseous necrosis [5]. The identification of the microorganism requires special stains (Ziehl-Nielsen). Culture of tissue biopsies on Lowenstein specific parts requires 4 to 6 weeks to obtain a result. Culture isolation of Mycobacterium tuberculosis is the gold standard for the diagnosis. It is essential because it is the only test that allows accurate identification of the organism and the realization of antibiograms, which is mandatory given the emergence of antibiotic resistance [20].

The tuberculin skin test (TST) is often difficult to interpret in our country since vaccination with Bacillus Calmette-Guerin (BCG) is widespread. It is often positive. It only has value if it is positive in an unvaccinated patient, or if it is phylectenular. However, in presence of any unilateral dragging symptoms, endoscopy with a nasopharyngeal biopsy is needed to remove an associated malignancy or a nasopharyngeal extension of tuberculosis infection, as was the case in our first patient. The differential diagnosis with other granulomatous diseases (leprosy, Wegener’s granulomatosis, sarcoidosis) depends on histopathological findings [3].

Since primary nasosinosal TB is extremely rare, medical investigations looking for other sites of TB infection is imperative. Diagnosis is based on culture on Lowenstein specific parts of different samples (nasal, sputum, gastric lavage, urine) [21]. In fact, Johnson and al. noted a generalized disease in over 75% of persons with nasal TB [8]. Treatment is based on anti-tuberculosis therapy [10, 13]. Conventionally, the drug association starts, during 2 or 3 months with four major antituberculosis drugs (rifampicin 10 mg/kg, Isoniazid 4-5 mg/kg, Pyrazinamide 20-30 mg/kg, Streptomycin 15 mg/kg, Ethambutol 15-20 mg/kg) [20].
After this initial phase, only Isoniazid-Rifampicin is continued for 4 to 6 months [5]. Sometimes, bacterial strains resist to one or all major anti-TB drugs. This often results from irregular or interrupted treatment before the required period. The chemotherapy is then required to be extended (18 to 24 month) and is more toxic to patients [13].

In a series of 50 cases of nasopharyngeal tuberculosis by Jian et al., all of the patients were disease-free after 2 years [22]. An associated surgery is classically recommended in the tumoral forms and in case of osseous invasion to repair bony defects, but, in the light of the rare published cases, it seems that, even in these cases, the antituberculosis chemotherapy used alone ensure the cure [13].

The disease progress is evaluated on the basis of several parameters: functional signs resolution, improvement of general conditions and normalization of biological and radiological assessments. Regular endoscopic control more or less associated with nasosinusal biopsy is the most important way to attest the good outcome and to allow stopping antituberculosis chemotherapy.

Prognosis is related to the type of lesions. Mucosal lesions are known to respond well to the regular treatment and usually have a favorable evolution. On the other side, the treatment is more difficult and the prognosis is worse in the presence of bone involvement or fistulas [14, 15]. This was the case in our third patient who presented an oro-antral fistula with a large palatal mucosa loss, and for whom, the total medical therapy period was of 12 months.

Nasosinusal tuberculosis remains a rare presentation of extra pulmonary tuberculosis. Its diagnosis is based on histological, clinical, radiological and histological findings. It usually arises from tuberculosis elsewhere justifying to search for other sites of the disease. This affection must be suspected in patient with chronic sinusitis resistant to medical treatment, especially when history of contact with infected individuals is related.

CONFLICT OF INTEREST

The authors declare that they have no conflict of interest.

REFERENCES


Haidar Hussien, et al