

Primary Submandibular Gland Tubercular Sialadenitis: A Case Report

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Abstract

Case Report

Tuberculosis of the submandibular gland is a rare pathology, the diagnosis of which is difficult, given the non-pathognomic clinical presentation and the absence of diagnostic guidelines, the portal of entry can be by hematogenous route or through the excretory canal of the gland. We present the case of a 33-year-old patient who presented with recurrent submandibular sialadenitis without explanation. In order to make a diagnosis a submandibulectomy was performed. The histological study showed a tubercular epitheliogigantocellular granuloma; the patient was put on antitubercular drugs for 6 months with a good evolution.

Keywords: Submandibular gland tuberculosis, histologic examination, tubercular epitheliogigantocellular granuloma, excisional biopsy.

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INTRODUCTION

Tuberculosis is a serious infectious disease, which is a public health problem in the world, especially in poor countries. Countries like India are considered endemic [1]. The increase in cases in developed countries is linked to HIV- AIDS and migration [2]. Despite its high frequency, it very rarely affects the major salivary glands, and is exceptional in the submandibular gland, this low incidence is explained by the resistance of these glands to mycobacterium tuberculosis [1, 3].

The head and neck location constitutes 15 to 20% of extrapulmonary tuberculosis cases, the most frequent location in the craniocervical region is the cervical nodes, followed by the larynx [4]. For the main salivary glands, the parotid location represents 70% of cases [5], followed by the submandibular location, the involvement of the sublingual gland has not been described [3].

We present a case of tuberculosis of the right submandibular gland, simulating recurrent sialadenitis in an immunocompetent patient.

OBSERVATION

A 33-year-old patient with a history of 3 episodes of right submaxillary sialadenitis, treated with

antibiotics with good evolution, presented to the maxillofacial emergency room of the Mohamed VI University Hospital in Marrakech, Morocco, with a subacute submaxillary swelling that had been evolving for 14 days, in a context of fever, without any notion of night sweats, or toothache, or alteration of the general condition.

The examination found a patient in good general condition, with a hot, red, painful and soft right submaxillary swelling. There were no palpable cervical adenopathies (figure 1).



Fig. 1: Installation of the patient on the operation table, swelling of the submandibular region

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Endobuccal examination found inflammation at the level of the ostium of the submandibular canal, pressure on the gland let out frank pus.

Ultrasound examination showed an infiltrated submandibular gland without any collection or detectable calculus. The cytobacteriological study of the pus showed no germ.

The patient was put under medical treatment based on bi-antibiotic therapy made of amoxicillin - clavulanic acid associated with metronidazole.

A biopsy-exeresis was decided upon due to the recurrence of the sialadenitis without explanation. The submandibulectomy was performed under general anaesthesia, by submandibular approach; the dissection was difficult and haemorrhagic due to the presence of numerous adhesions (figure 2 et 3).



Fig. 2: Sub-mandibulectomy



Fig. 3: Surgical specimen

The postoperative course was simple.

The anatomopathological study of the submandibulectomy specimen showed an epitheliogigantocellular granuloma with central caseous necrosis in favor of tuberculosis.

The patient was referred to the department of phthisiology, where the examination denied the presence of other localizations, thus confirming the primary character of the localization in the right submandibular gland.

The patient was put under the therapeutic protocol adopted in Morocco for tuberculous adenitis; this protocol is made of the association of isoniazid, rifampicin, ethambutol and pyrazinamide (HREZ) for two months, then isoniazid and rifampicin (HR) four months.

The evolution under treatment was good.

The 1-year check-up found a patient in good general condition, with a notion of weight gain, the physical examination showed a depression in the right submandibular region, without masses or palpable lymphadenopathy (figure 4).



Fig. 4: Follow up at 1year, submandibular region depression

DISCUSSION

According to the WHO 2020 report, tuberculosis was the 13th leading cause of death in the world. 10.6 million people are estimated to be newly

infected in 2021, with a total of 1.6 million deaths in 2021, of which 187,000 will be from HIV infection. An increase of 3.6% was noted between 2020 and 2021. 86% of cases were diagnosed in 30 countries, with two-thirds of these cases concentrated in 8 countries, with India (28%) in first place, followed by Indonesia (9.2%) and China (7.4%) in third place [6].

The head and neck location constitutes 10% of extrapulmonary tuberculosis cases. Tuberculosis of the major salivary glands is rare, and it is exceptional in the submandibular gland, only a few cases are published. This is due to the resistance of these glands to mycobacterium tuberculosis, this resistance can be explained by the presence in the salivary secretion of thiocyanate ions and proteolytic enzymes, associated with the continuous salivary flow of these glands.

Tuberculosis of the salivary glands may be primary or secondary. The secondary form occurs following dissemination from a distant site such as the lung, by hematogenous or lymphatic route, or by dissemination from a nearby site in the oral cavity such as a tonsil or a tooth. The primary form is rarer, and most often involves the parotid gland.

The clinical picture can be misleading, the general constitutional signs of tuberculosis are usually absent, such as fever, asthenia, loss of weight and appetite [3, 7, 8]. This form of tuberculosis can simulate a tumor by its progressive evolution, or a chronic sialadenitis, which was the case of our patient. It can even coincide with the presence of a calculus in the salivary duct, which makes the diagnosis even more complicated. Most often the disease starts with a progressive and painless swelling of the gland [4]. An acute installation is possible, in the form of a swelling under angulomandibular, with severe pain, most often this form is diagnosed as a dental abscess [8], bilateral involvement of the submandibular gland has been described only once [9].

Clinically, there is a firm, non-fluctuating, fixed mass in the submandibular region without inflammatory skin signs. The presence of cervical adenopathy is possible but not mandatory. The endobuccal examination is crucial, it evaluates the state of the oral floor, notably Wharton's canal and salivary secretion (clear or purulent), and also the state of the teeth in search of caries [3].

Delayed diagnosis is typical of tuberculous sialadenitis, given its rarity. This diagnosis is even more difficult when it is a primary submandibular tuberculous sialadenitis. Most often the diagnosis is confirmed by histopathological study on a submandibulectomy specimen [3, 10, 11].

Imaging has only a diagnostic orientation role. Ultrasound is the first-line examination, it visualizes a

swelling of the gland with an altered echostructure. It can also show foci of glandular and/or lymph node necrosis.[10]. Given the limitations of confirming the nature of the lesion [3]. This examination allows detection and description of lesions larger than 5 mm in diameter. The gland is swollen, with thickening of the cutaneous soft parts and thinning of the fatty planes, the caseous necrosis appears as hypodensity [8]. The most suggestive CT appearance of tuberculous submandibular sialadenitis is that of multiple, round, smooth-walled lesions with central glandular hypodensity and enhanced parenchyma [12, 13]. However, the CT scan cannot distinguish between a tubercular origin and a tumoral origin of the lesions. MRI is a very efficient but very expensive examination, the MR sialogram is essentially reserved for the study of salivary gland ducts [3, 13].

Diagnostic confirmation can be performed by several methods with different sensitivity and specificity, fine needle aspiration cytology, acid-fast staining with culture, PCR and incisional or excisional biopsy [3]. Fine needle aspiration cytology (FNAC) is a minimally invasive and safe procedure and is considered by some authors to be the examination of choice for diagnosing tuberculosis of the submandibular gland [1, 3]. Studies have shown that the sensitivity of the FNAC is higher than 80% and its specificity between 94% and 100% [1, 4], the diagnostic accuracy is 33.3% according to Kim *et al.*, [14].

BAAR staining associated with culture has a sensitivity of less than 29%, low for the diagnosis of submandibular tuberculosis. This sensitivity increases to 85% if the culture is associated with the FNAC [10]. In addition, the combination PCR/FNAC can increase the sensitivity and specificity to 90% [1, 7, 8, 15]. PCR is a quick and easy test; however, it remains expensive, inaccessible in our context, and a source of false positives [16, 17].

Given the risk of chronic fistula, incisional biopsy is contraindicated [8]. Surgical excision is justified by diagnostic impasse using the examinations described above [3, 18, 19].

The coexistence of tuberculous sialadenitis with a malignant tumor has been described, but remains rare [20, 21].

Given the rarity of this location of tuberculosis, no protocol is proposed by the WHO, the protocol described in the literature is the one that starts with a combination of 4 molecules (isoniazid, rifampicin, pyrazinamide and ethambutamol) for a duration of 2-4 months, followed by a combination of 2 molecules (isoniazid and rifampicin) for a duration of 6-12 months [1, 3, 17].

Surgical treatment is indicated in case of failure or non-response to medical treatment, in case of morphological repercussions of the swollen gland, in case of failure of other tests to confirm the diagnosis, and/or in case of complications [1, 3]. In our case, the surgical treatment was decided in front of the diagnostic impasse.

Anti-tuberculosis medical treatment must be continued after surgical removal of the gland [1].

CONCLUSION

Tuberculosis of the major salivary glands is a rare condition, exceptional in the submandibular gland. Submandibular tuberculosis sialadenitis, which is exceptional, is essentially manifested by a swelling of the submandibular gland associated or not with general signs (fever, night sweats, AEG). Diagnosis is based on bacteriological (presence of BAARs on FNAC, PCR) or histological (on submandibulectomy specimen) confirmation. Anti-tuberculosis treatment remains the primary treatment, whether or not submandibulectomy is performed.

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