Case Report

Granuloma of the labial minor salivary glands in tuberculosis

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Abstract

Minor salivary gland granuloma is rare in tuberculosis. We present a case of labial minor salivary gland granulomas occurring in the course of tuberculosis along with skin, mediastinal and liver granulomas. The patient (46-year-old man) presented with asthenia, nocturne transpiration, weight loss and compressive thoracic adenopathies. The angiotensin I converting enzyme was elevated as well as calcemia. The QuantiFERON test and culture of a mediastinal specimen were positive for *Mycobacterium tuberculosis*. Multinucleated-cell-granulomas (focally with necrosis) were identified on skin, mediastinal, liver and minor salivary gland biopsies. Kidney biopsy was suggestive of IgA-glomerulonephritis. Treatment (isoniazid, rifampicin, ethambutol and moxifloxacin) was started and corticoids 15 days afterward. At 1 year, the patient had recovered. In conclusion, a case of labial minor salivary gland granulomas occurring in the course of tuberculosis is reported. Tuberculosis should be included in the differential diagnoses of labial minor salivary gland granulomas as treatments may be more complex than those for other granulomas including sarcoidosis.

Keywords: Granuloma, labial, minor salivary gland, sarcoidosis, tuberculosis

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INTRODUCTION

Labial/minor salivary gland granulomas occur more frequently in the context of sarcoidosis.[1,2] However, in a recent series coming from France, epithelioid and giant cell granulomas were found in the labial salivary glands of 7 out of 65 patients with tuberculosis. In all cases, there were no labial or salivary gland abnormality. The main feature was microscopic, consisting in the lack of caseating necrosis in all the studied biopsies. The favorable response to antituberculosis treatment allowed to rule out an associated sarcoidosis.[2]

Here, we present a case of labial minor salivary gland granulomas occurring in the course of tuberculosis along with skin, mediastinal and liver granulomas.

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CASE REPORT

A 46-year-old man presented with asthenia, nocturnal transpiration, weight loss (17 kg/3-months), dysphagia and compressive thoracic adenopathies (absent on the initial radiography and present on the computed tomography-scan 3-month afterward). Blood tests showed hepatic cytolysis and cholestasis. Serologies were negative for C-hepatits, HIV, Epstein–Barr, Parvo, HHV8 viruses and for bilharzias and leishmania. The serum angiotensin I converting enzyme (ACE) was elevated (88, normal value <68 IU/L) as well as calcemia, creatininemia and uremia. The QuantiFERON test and culture of a mediastinal compressive adenopathy specimen were positive for *Mycobacterium tuberculosis*. Granulomas were noted on the skin, mediastinal (with significant necrosis)

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and liver and minor salivary gland biopsies. Kidney biopsy was suggestive of IgA-glomerulonephritis. Treatment with isoniazid, rifampicin, ethambutol and moxifloxacin was started. Corticoids were added 15 days afterward. A labial minor salivary gland biopsy was made due to persistent hypercalcemia at 14 days after the beginning of antituberculosis treatment. The glandular lobules showed foci of lymphocytic infiltrate with several polymorphonuclear neutrophils and several epithelioid and giant cell granulomas, some pericanalar with edematous and incipient necrosis [Figure 1a and b]. The Ziehl–Neelsen stain did not show acid-alcohol-resistant bacilli (AARB). At 26 days after the beginning of the treatment, the patient showed no clinical complaints and calcemia was normal; at 1 year, the patient had recovered.

DISCUSSION

Here, we present a rare case of labial minor salivary gland granuloma occurring in the initial course of tuberculosis. Salivary gland tuberculosis has been reported in major salivary glands such as parotid and submandibular glands as well as in minor salivary glands.[2-4] The main differential diagnosis, both clinical and histological, in adults is sarcoidosis. A cytological examination may be useless since features of granulomas are nonspecific although polymorphonuclear leukocytes may be observed.[5] The histological diagnosis may also be rendered difficult by the lack of necrosis and by the absence of AARB on the Ziehl–Neelsen-stained tissue section. However, necrosis is reported to lack in minor salivary gland granulomas from patients with tuberculosis, isolated, or supposed associated to sarcoidosis.[2,6,7]

One may question if the two diseases, tuberculosis and sarcoidosis, coexisted in the patient we report. This is suggested by reported data stating that minor salivary gland granulomas do not occur in tuberculosis and that in those rare cases of minor salivary gland granulomas the two diseases could have been in fact associated.[1] Skin granulomas are unusual in tuberculosis, however reported.[2] Elevated ACE may occur in both affections. Moreover, IgA-nephritis, as seen in the present case, may be associated with elevated ACE.[8] We did not identify sarcoidosis-specific cellular inclusions or Schaumann bodies. Moreover, normalization of calcemia after beginning of treatment might favor the hypothesis of tuberculosis-related granuloma rather than sarcoidosis-related granuloma. Hypercalcemia associated to multiorgan granulomas may explain clinical renal complaints. In the case we report, there was no granulomatous nephritis but IgA-glomerulonephritis, such an association being already reported not only in sarcoidosis but also in two patients with pleuropulmonary tuberculosis.[9-11]

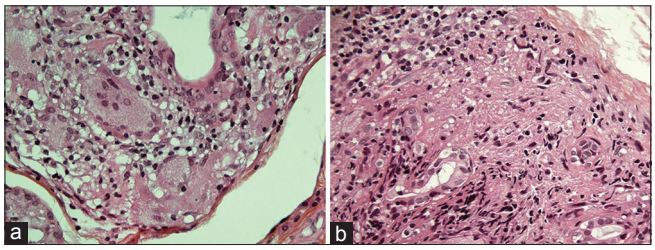

Figure 1: (a) The salivary gland lobules showed epithelioid and multinucleated giant cell granulomas (H&E, ×40) (b) Focal necrosis was observed (H&E, ×40)

In conclusion, labial minor salivary gland granulomas may occur in the course of tuberculosis with mediastinal, liver and skin localizations. Associated sarcoidosis cannot completely be ruled out although of limited clinical relevance as suggested by the posttreatment resolutive evolution of disease in the presented case.

Declaration of patient consent

The author certifies that all appropriate patient-related consent forms have been fullfilled in order to protect the patient's rights. The manuscript does not contain patient photographs, names (or initals) neither race, country or continent informations nor identifiable clinical or paraclinical data.

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Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Tabak L, Ağirbaş E, Yilmazbayhan D, Tanyeri H, Güç U. The value of labial biopsy in the differentiation of sarcoidosis from tuberculosis. Sarcoidosis Vasc Diffuse Lung Dis 2001;18:191-5.
2. Delcey V, Morgand M, Lopes A, Mouly S, Jarrin I, Sellier P, *et al.* Prevalence of granulomatous lesions in minor salivary gland biopsy in a case series of 65 patients with tuberculosis. Rev Med Interne 2016;37:80-3.
3. Christoforidou A, Kyrgidis A, Markou K, Triaridis S. Radiology quiz case 1. Tuberculosis of the right parotid gland. JAMA Otolaryngol Head Neck Surg 2013;139:187-8.

4. Tauro LF, George C, Kamath A, Swethadri G, Gatty R. Primary tuberculosis of submandibular salivary gland. J Glob Infect Dis 2011;3:82-5.
5. Kaur G, Dhamija A, Augustine J, Bakshi P, Verma K. Can cytomorphology of granulomas distinguish sarcoidosis from tuberculosis? Retrospective study of endobronchial ultrasound guided transbronchial needle aspirate of 49 granulomatous lymph nodes. Cytojournal 2013;10:19.
6. Milleron B, Herman D, François T, Favre M, Gauthier JM, Akoun G, *et al*. Biopsy of labial accessory salivary glands and tuberculosis. Nouv Presse Med 1982;11:1337.
7. Nessan VJ, Jacoway JR. Biopsy of minor salivary glands in the diagnosis of sarcoidosis. N Engl J Med 1979;301:922-4.
8. Mizuiri S, Hemmi H, Arita M, Aoki T, Ohashi Y, Miyagi M, *et al*. Increased ACE and decreased ACE2 expression in kidneys from patients with IgA nephropathy. Nephron Clin Pract 2011;117:c57-66.
9. Bergner R, Hoffmann M, Waldherr R, Uppenkamp M. Frequency of kidney disease in chronic sarcoidosis. Sarcoidosis Vasc Diffuse Lung Dis 2003;20:126-32.
10. Singh P, Khaira A, Sharma A, Dinda AK, Tiwari SC. IgA nephropathy associated with pleuropulmonary tuberculosis. Singapore Med J 2009;50:e268-9.
11. Ortmann J, Schiffl H, Lang SM. Partial clinical remission of chronic IgA nephropathy with therapy of tuberculosis. Dtsch Med Wochenschr 2010;135:1228-31.