CASE REPORT

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Mycobacterial spindle cell pseudotumor (MSP) of the nasal septum clinically mimicking Kaposi's sarcoma: case report*

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SUMMARY

Mycobacterial spindle cell pseudotumor (MSP) is a rare benign lesion characterised by local proliferation of spindle – shaped histiocytes containing acid – fast atypical mycobacteria, clinically resembling Kaposi`s sarcoma. Most cases of MSP reported so far affected immunodeficient patients or patients receiving immunosuppressive medication.

We report a case of MSP affecting the nasal septum of a 76-year-old man, a location that has not been published so far concerning the manifestation of MSP.

In conclusion, our case report points toward MSP as a very rare pseudomalignant lesion that should be included into deliberations concerning the differential diagnosis of circumscribed expansile nodular proliferations of the nasal septum clinically mimicking Kaposis`s sarcoma or other mesenchymal neoplasms.

Key words: nasal septum, pseudomalignant mesenchymal lesions, mycobacterial spindle cell pseudotumor (MSP), atypical mycobacteria

CASE REPORT

A 76-year-old man presented with a firm nodular mass lesion affecting the right nasal septum which was discovered incidentally without causing any symptoms. Additionally, he suffered from hypertension treated with Losartan (50mg daily) and from prostate cancer which was treated by local radiation because the patient refused any surgical procedures. There was no evidence of any lymphatic or distant metastases at presentation.

Physical examination showed a well circumscribed expansile nodule underneath a superficially intact mucosa of the right nasal septum clinically resembling Kaposi`s sarcoma. The lesion was extirpated under local anaesthesia. The resected specimen measured 5 x 4 x 3 mm.

Histological examination revealed a well circumscribed nodule composed of clusters of spindle – shaped cells admixed with a sparse inflammatory infiltrate, covered by intact multilayered squamous epithelium (Figure 1). Immunohistochemistry was performed to better define the cell population comprising the lesion. The spindle cells stained strongly with anti-CD68 (Figure 2) indicating a monocytic / histiocytic origin. Mitotic activity was assessed by immunostaining for Ki67 and was estimated to be less than 1%. The cytoplasm of the spindle cells contained Periodic – Acid – Schiff (PAS) positive bacilli, which turned out to be acid – fast on subsequent Ziehl – Neelsen stain. This finding was felt to be consistent with atypical mycobacteria. However, cultures performed on a portion of the specimen using paraffin – embedded tissue were negative. Polymerase chain reaction (PCR) was performed as described by Soini et al. [1], yielding a 423 bp amplification product (Figure 3). Because the lesion was too small to provide any further samples for analysing, the mycobacterial subspecies could not be determined. The patient underwent clinical, radiographic, and laboratory screening. Routine hematology and chest radiograph were

Figure 1. Nodular lesion composed of spindle – shaped cells admixed with a sparse inflammatory infiltrate, covered by intact multilayered squamous epithelium. Hematoxylin and eosin, x10.

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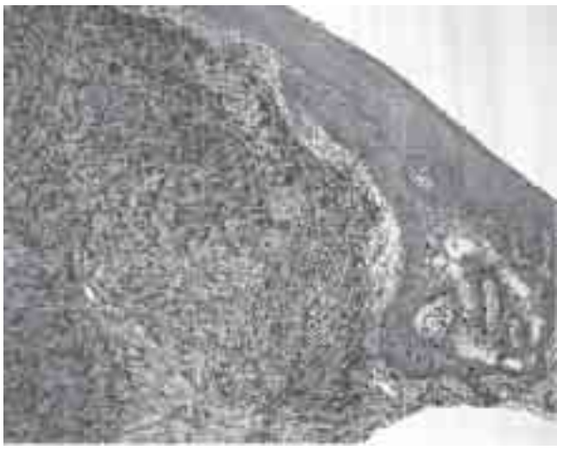

Figure 2. Immunostaining for CD68 highlights the presence of spindle – shaped histiocytes. anti – CD68, x10.

Figure 3. Ethidium bromide stained 1,5 % agarose gel. Lanes: I = 100 bp DNA marker, II = patient`s sample, III= negative control.

normal. Intradermal skin testing using the Mantoux test was negative. No mycobacteria were isolated in cultures from blood, sputum and stool specimens. Serological investigations and HIV testing were negative. There was no evidence of any profoundly suppressed immune status, especially CD4/CD8 ratio was found within normal limits. Administration of any immunosuppressive medication in the past was denied by the patient. Because there was no evidence of any systemic manifestation, no antimicrobial pharmacotherapy was initiated. There was no evidence of any local recurrence or of any other manifestations of the „disease" at the eight - month postsurgical follow – up.

DISCUSSION

MSP are rare lesions characterised by local proliferation of spindle – shaped histiocytes caused by atypical mycobacterial infection. The first case of MSP was reported by Wood et al. occuring in the skin of an immunosuppressed cardiac transplant recipient [2]. Clinically, MSP mimicks Kaposi`s sarcoma or other mesenchymal neoplasms [3]. In the past, a few cases have been reported affecting lymph nodes in HIV positive patients [3,4]. Atypical mycobacterial infections are a well known frequent problem in immunodeficient patients. The great majority is caused by Mycobacterium avium complex. However, even in immunodeficient patients, MSP does not necessarily present with multivisceral involvements. Morrison et al. reported a case of MSP limited to the brain in a patient receiving steroid therapy for treatment of sarcoidosis [5].

The presence of potentially pathological atypical mycobacteria which are practically ubiquitarious does not correspond to „infection", because they are also found in nasal secretions of healthy individuals [5]. There is no evidence of person – to – person transmission [6,7].

Generally, the diagnosis of MSP should prompt further workup to rule out any underlying immunodeficiency. Especially, serological testing for HIV should be performed. Concerning clinical management, any systemic or multivisceral spread of atypical mycobacteria requires sufficient pharmacotherapy. Nontuberculous mycobacterial infections are difficult to treat and do not respond to traditional antituberculous agents. However, antimicrobial combinations that include clarithromycin or azithromycin have been shown to be effective in treating several nontuberculous mycobacterial infections [7]. In conclusion, MSP should be kept in mind as a possible differential diagnosis when evaluating expansile nodular lesions affecting the nasal septum, especially in immunodeficient patients or in patients receiving immunosuppressive medication.

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