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CASE REPORT

Cytological Diagnosis In A Case Of Histoid Leprosy

CHUFAL S S*, MANNAN R**, SINGH M***, YADAV S****

ABSTRACT

Histoid leprosy, a variant of lepromatous Leprosy, usually develops due to resistance to dapsone monotherapy. It also can arise denovo. The clinical presentation of Histoid leprosy can mimic other inflammatory and some neoplastic conditions, especially if it arises denovo. FNA cytology can be performed as an outdoor procedure and can provide a quick and reliable diagnosis in such cases. A 35 year old male presented with multiple nodules all over the body. There was no previous history of Leprosy or antileprotic treatment. The clinical differential diagnoses were Post Kala azar dermal Leishmaniasis (PKDL), Histoid Hansen's leprosy and Cutaneous metastasis. FNA with a 23G needle showed numerous histiocytes which were filled with intracellular and extracellular negative bacillary images. These bacilli were arranged parallel to each other rather than in a random manner, which differentiated it from Atypical mycobacterial cutaneous infection, although a rare condition, without the aid of special stains and other ancillary investigations. Ziehl Neelsen's staining for acid fast bacilli was positive. The arrangement of the negative bacillary images was important in distinguishing it from Atypical Mycobacterial infection, as FNA smears can be similar in both the cases and the treatment for both the entities is different. Subsequent histopathology confirmed the diagnosis and the patient responded to the treatment.

Keywords- Histoid Leprosy, Negative Bacillary images, Atypical Mycobacterial infection

Key Message:

- FNA cytology is a simple, safe and a least traumatic procedure which can provide a quick and reliable diagnosis in cases with histoid leprosy.
- FNA helps in ruling out other probable clinical differential diagnoses in de novo histoid patients, so that treatment can be started at the earliest, as such patients can form potential reservoirs of infection.
- Cutaneous Atypical mycobacterial infection should also be considered in clinical differential diagnoses in such de novo histoid patients, as FNA smears can be similar in both the cases.
- The arrangement of negative bacillary images within the cytoplasm of the histiocytes is important in distinguishing the de novo histoid patients from atypical mycobacterial infections without the aid of special stains or other specific tests.

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Introduction

Hansen's disease (Leprosy) is a chronic slowly progressive infection which is caused by *Mycobacterium Leprae*, affecting the skin and the

peripheral nerves, resulting in disabling deformities. The prevalence of leprosy in India is reported to be 0.7 per 10,000 Population [1]. The disease manifests itself in two forms, namely lepromatous leprosy

and tuberculoid leprosy, lying at the two ends of a long spectrum of disease. Histoid Leprosy is a variant of Lepromatous Leprosy, which mostly develops due to resistance to dapsone monotherapy, although it can also arise denovo[2]. The incidence of histoid hansen's disease has been described to be 2.79 – 3.60 % in Leprosy patients [3] The cases of denovo histoid patients are on the rise in India [4]. Here, we report a case of 35 year old patient who had multiple subcutaneous nodules all over the body including face, without any previous history of leprosy or antileprotic treatment, causing a diagnostic dilemma clinically. FNA cytology was performed and a provisional diagnosis of Histoid Leprosy was given, which was later confirmed by histology. On the FNA smears, the arrangement of negative bacillary images within the histiocytes is important to differentiate histoid hansen's disease from cutaneous atypical mycobacterial infections.

Case Report

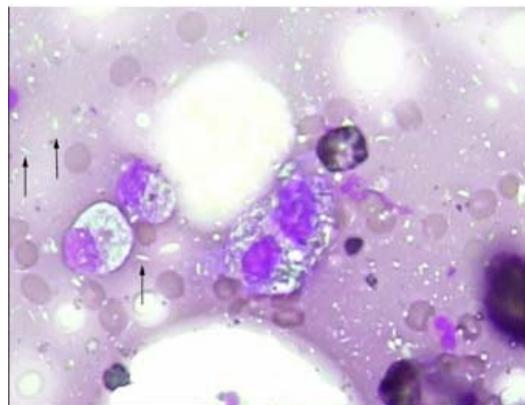
A 35 year old male presented with multiple, skin coloured, subcutaneous juicy nodules and ulcers all over the body, including the face. [Table/Fig1] The clinical differential diagnoses were Histoid Hansen's disease, Post Kala azar dermal Leishmaniasis and cutaneous metastasis. There was no previous history of leprosy or anti leprotic treatment. The lesions started as a single ulcerated growth over the inner aspect of the thigh, with gradual eruption of these lesions all over the body.



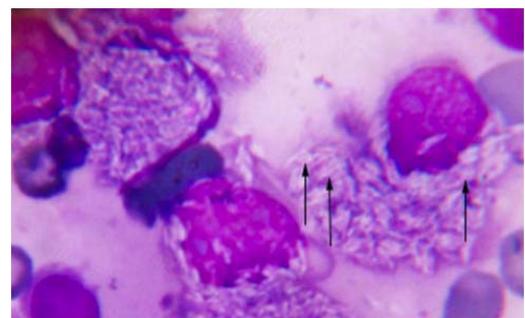
[Table/Fig 1]: Clinical picture of the patient showing multiple subcutaneous nodules all over the body

Three consecutive night peripheral blood smears showed no evidence of haemoparasites including microfilarae. The patient's serological tests for VDRL and HIV were negative, Mantoux's test was

negative, the aldehyde test for Leishmaniasis was negative and other ancillary investigations like Chest X- ray, USG abdomen and careful physical and clinical examinations were unremarkable. FNA with a 23G needle, with multiple punctures from four different lesions yielded blood mixed material. The smears which were prepared by staining with May Grunwald's Giemsa, showed numerous foamy histiocytes which were filled with intracellular and extracellular negative bacillary images.[Table/Fig.2] These histiocytes were admixed with few spindle shaped cell clusters. The negative bacillary images were arranged in a parallel disposition, rather than in a random manner within the histiocytes.[Table/Fig.3]. This arrangement of bacilli ruled out the other rare cutaneous infection which is caused by Atypical mycobacteria, having a similar picture cytologically.



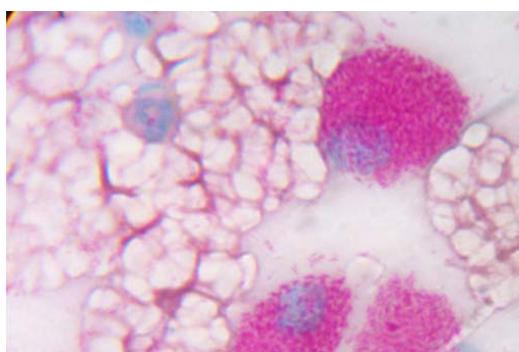
[Table/Fig 2]: Cytological smears showing histiocytes with negative bacillary images intracellularly extracellularly. (MGG 100x)



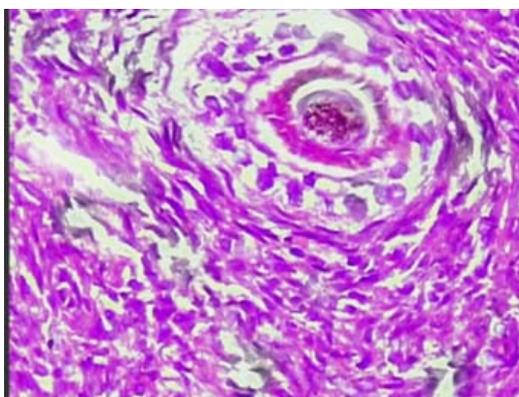
[Table/Fig 3]: Cytological smears showing parallel disposition of negative bacillary images with in Histiocytes.(MGG 400x)

Ziehl Neelsen's staining for acid fast bacilli showed numerous solid staining bacilli in clumps within the histiocytes, as well as lying extracellularly. [Table/Fig.4]. Taking into account the clinical

presentation and the cytological findings, especially the arrangement of the negative bacillary images in the parallel disposition and the presence of acid fast bacilli, a diagnosis of histoid hansen's disease was given. Subsequently, a punch biopsy of the lesion was done and histopathology revealed numerous spindle shaped macrophages which were arranged in a storiform pattern beneath an ulcerated epidermis with a Grenz zone in between. These spindle cells were also arranged around the skin adnexa, as if compressing them. [Table/Fig.5]. Thus, a diagnosis of Histoid Leprosy was confirmed and the patient responded to the treatment.



[Table/Fig 4]: Extracellular and intracellular Acid Fast Bacilli. (ZN 1000x)



[Table/Fig 5]: Histological section showing spindle shaped macrophages around skin adnexa.(H&E 400x)

Discussion

Although Leprosy is a common problem in India, in recent years, its incidence has declined and the pure histoid forms are not so common, although the incidence of denovo histoid patients is increasing [4]. Histoid Leprosy occurs predominantly in male patients who take irregular treatment for Leprosy [3] or it arises denovo in Lepromatous Leprosy patients [2]. It also has been described in patients who develop de novo Histoid Leprosy during the course

of the DEC treatment for filariasis [5],[6]. The patient failed to give any history of leprosy or antileprotic or antifilarial therapy. Three consecutive night peripheral blood smears showed no evidence of haemoparasites including microfilarae. Histologically it mimics Dermatofibromas, whereas clinically it may resemble Post Kala azar dermal Leishmaniasis (PKDL) and in some cases, nodular syphilis [7], eruptive Keratoacanthomas or cutaneous metastasis [3]. However, the patient's serological tests for VDRL and HIV were negative, the aldehyde test for Leishmaniasis was negative and other ancillary investigations like Chest X- ray, USG abdomen and careful physical and clinical examinations showed no evidence of malignancy. FNA from four different sites was performed, as it is important to perform multiple punctures to yield cellular material [8]. The smears showed numerous spindle to oval histiocytes which were filled with numerous negative bacillary images, as well as those which were lying extracellularly [Table/Fig 2, 3]. Such a picture can be seen in Cutaneous Atypical Mycobacterial Infections, although rarely [9]. The arrangement of the negative bacillary images was important in distinguishing it from Atypical mycobacterial infections, although described in lymph nodes [10],[11]. These bacilli were arranged in a parallel disposition, whereas in Atypical mycobacterial infections, they are arranged randomly [12]/[Table/Fig3]. As the patient had no past history of leprosy or antileprotic treatment and as both forms of infections can be seen in immunocompromised patients in a tropical country like India, it was important to distinguish such a de novo histoid Leprosy patient, as the treatment for both these conditions was different. Strong Acid Fast bacilli positivity on cytology and the characteristic histology confirmed the diagnosis. Thus, it can be concluded that FNA is a rapid and cost effective way of detecting mycobacteria, as well as in differentiating Histoid Leprosy patients from cutaneous Atypical mycobacterial infections which can present with similar clinical pictures, without the aid of special stains and other specific tests [13]. The case highlights the role of cytology as a quick, easy and a minimally invasive outdoor procedure to rule out other conditions, relieve the patient's anxiety and to help the clinician to start the appropriate treatment at the earliest [14]. As the bacillary load in such Histoid patients is very high, they can be potential reservoirs of infection [4]. Other investigations like histopathology may not be

acceptable to the patient, cultures and PCR can be time consuming, and the slit smear technique stained with Ziehl Neelson's stain is limited to the determination of the presence or absence of Acid Fast bacilli[8].

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