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Case Report

Histoid Hansens with Multiple Nerve Abscesses in a Treatment-naïve Patient in Post-Elimination Era

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Histoid Hansen is a variant of multibacillary disease leprosy, that was earlier seen following dapsone monotherapy or resistance and of late noted de-novo. It is believed to have focal loss of immunity. We report a de-novo case of Histoid Hansen with severe type 1 reaction with multiple nerve abscesses, such presentation has not yet been reported in literature. Patient responded well to multibacillary multidrug therapy along with oral prednisolone, he did not show signs of any sensory/motor loss and in two weeks, subsiding nerve tenderness and flattening of the skin lesions was noted. As per earlier experience published in literature once an abscess is formed with caseous necrosis within the nerve sheath it does not resolve by medication alone. Follow up of such cases by high-resolution ultrasound (HRUS) and or confirmation by wedge biopsy is needed to confirm the same.

Keywords: Histoid, Hansens, Leprosy, Nerve Abscess, Type 1 Reaction, Lepra Reaction

Introduction

Histoid Hansen is a variant of a multibacillary disease leprosy first described by Wade. Dome shaped papules are noted on normal appearing skin which consist of spindle shaped cells in storiform pattern on histology. A focal loss of immunity leading to its formation is postulated (Kaur et al 2009). Type 1 lepra reactions are less often encountered in Histoid Hansen's patients Dhattarwal et al 2023, Singh et al 2014/ 2015, Sun et al 2017), none reported nerve abscess. We report a patient with Histoid Hansen with de-novo presentation with multiple nerve abscesses.

Case report

A 31-year-old male presented with multiple asymptomatic skin-colored and erythematous shiny papules over apparently normal skin over the forehead, bilateral pinna and bilateral arms (Figs 1a, 1b) for 2 years along with painful swellings on the bilateral lower limbs for 6 months. These nodules were firm and tender, measuring approximately 3cm x 2cm, noted along the cutaneous nerves i.e. the right sural nerve, right anterior tibial nerve, and bilateral posterior tibial nerve (Figs 2a, 2b). Bilateral ulnar nerves, common peroneal nerves, and posterior tibial nerves and radial cutaneous nerves were

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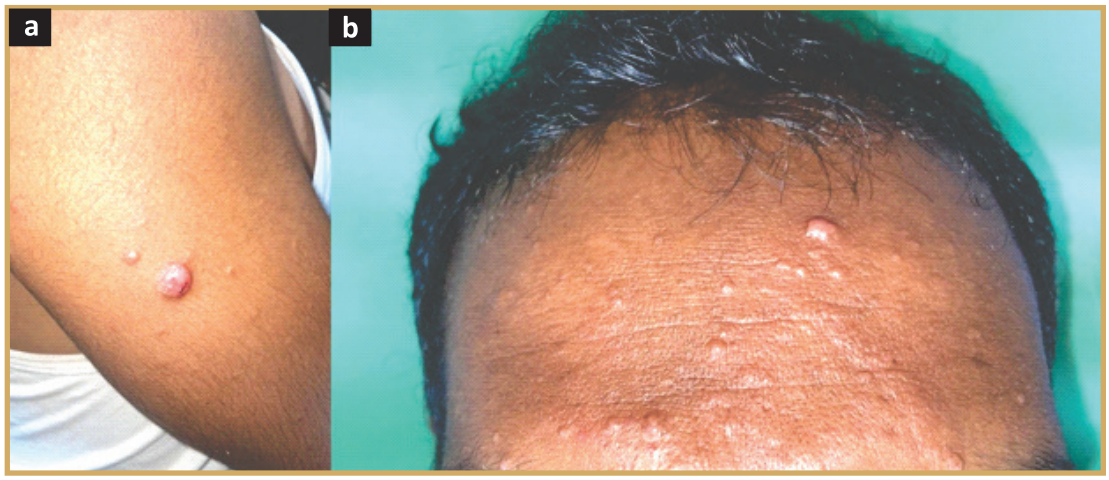

Fig. 1a : Classical erythematous to fleshed colored nodule of a histoid leprosy on right arm.
Fig. 1b : Multiple, discrete erythematous skin-colored papules on forehead.

Fig. 2a : Multiple swellings (arrow) along cutaneous branches of superficial peroneal nerve.
Fig. 2b : Swelling along right sural nerve with xerotic changes on the skin.

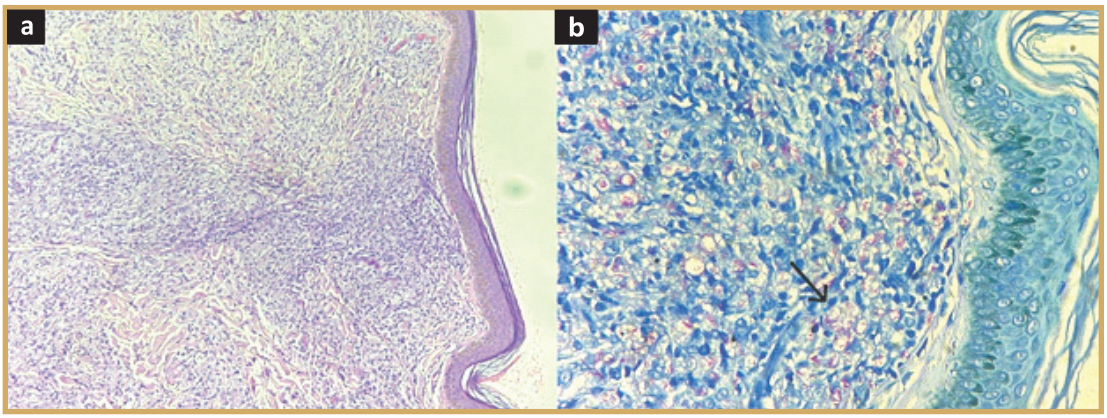

Fig. 3a: Numerous spindle-shaped non-vacuolated histiocytes arranged in interlacing bands in a whorled pattern. (Hematoxylin and Eosin stain, 40X).

Fig. 3b : Foamy macrophages with Fite staining positive for acid-fast bacilli (100X). (arrow)

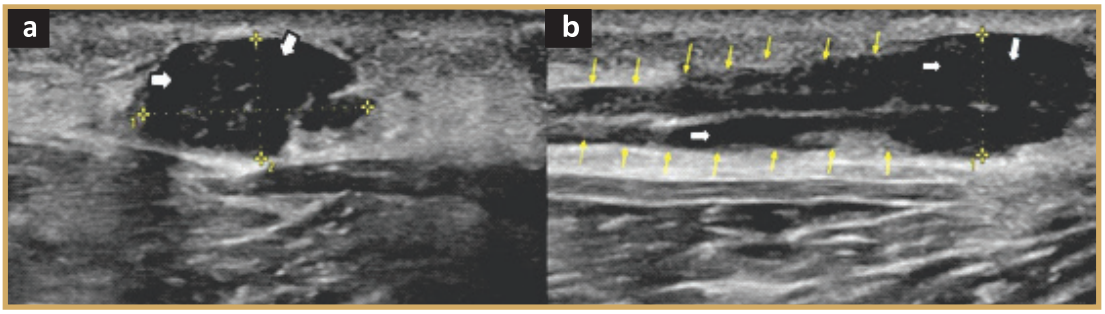

Fig. 4a : High resolution ultrasound (axial view) of the right common peroneal nerve (denoted by the yellow dotted lines & plus sign) depicting diffusely thickened right common peroneal nerve with hypoechoic areas within (denoted by the white arrows) suggestive of nerve abscess.

Fig. 4b : High resolution ultrasound (longitudinal view) of the left common peroneal nerve (outlined by the yellow arrows)– depicting diffusely thickened right common peroneal nerve with hypoechoic areas within (denoted by the white arrows) suggestive of nerve abscess.

symmetrically thickened. No sensory or motor deficits were noted. He denied prior anti-leprosy treatment. We considered a differential of Histoid Hansens with nerve abscess or neural histoid. Slit-skin smear from papules on the forehead and ear lobes showed bacteriological indices of 6+. Histopathology from a papule on the right forearm confirmed Histoid Hansen (Fig 3a, 3b) and high-resolution ultrasound (HRUS) of swellings and nerves suggested abscesses (Fig 4a, 4b).

Considering the clinical, histopathological and HRUS findings, a diagnosis of histoid Hansens with Type 1 reaction with nerve abscess was arrived at.

Patient was started on multibacillary multidrug therapy along with oral prednisolone (0.75mg/kg body weight) as he did not show signs of any sensory/motor loss. In two weeks, he noted subsiding nerve tenderness and flattening of the skin lesions. The possible need for epineurotomy was discussed in case of recurrence of the swelling on the nerve. As there was rapid decrease in the size of the nerve abscess following the steroids, hence we continued the steroids and the patient chose to go to hospital close to his hometown as per his convenience. No follow-up HRUS could be done.

Discussion

Histoid leprosy is a rare variant of lepromatous leprosy characterized by distinct morphological and histopathological appearance. Though originally described in patients taking dapsone monotherapy, it is now common in treatment-naïve patients similar to this patient (Kaur et al 2009, Mendiratta et al 2011).

Leprosy reactions are rarely reported in Histoid Hansen, with type 2 reactions being more common than type 1 as it is multibacillary. There are 3 case reports with documented type 1 reaction with varied presentations of erythematous skin lesions, ulceration, and neuritis (Dhattarwal et al 2023, Singh et al 2014/2015, Sun et al 2017) However, none have reported nerve abscess. Single documented report of nerve abscess is noted along with Type 2 lepra reaction in Histoid Hansen (Kaur et al 2009, Mendiratta et al 2011, Patvekar et al 2014). In our patient, the lesions were erythematous with neuritis and multiple nerve abscess. The rapid response to steroids with flattening of lesions further reaffirms the diagnosis of type 1 reaction.

Though Histoid Hansen is considered a variant of lepromatous leprosy, enhanced immune response including cell-mediated immunity as well as humoral immunity is noted against *Mycobacterium leprae* when compared with lepromatous leprosy. Also, it is also postulated that there is lack in the function of macrophages rather than deficiency leading to inability to kill bacilli.

In Histoid Hansen, studies on immuno-histochemistry showed decreased dendritic cell activity on one hand and on the other, CD4, CD8 and CD 6 were similar to that in tuberculoid leprosy (Kontochristopoulos et al 1995). CD3+, CD20+ and CD 8+ in the histoid lesions were in higher number (Da Costa et al 2013). Hence some authors do not agree to interpret that Histoid Hansen is part of lepromatous spectrum (Kaur et al 2009).

In patients with type1 reaction in leprosy, predominant CD4 + T lymphocytes with Th1 cytokines (IFN-γ, IL-2, IL1, IL-6, IL-17) are noted, whereas in erythema nodosum leprosum, Th2 response (IL-2, IL-4, IL-5, IL-6, and IL-10 cytokines) predominates.

Nerve abscesses in leprosy are typically 'cold' and painless, often can be mistaken for a soft tissue tumour or nerve tumour or rarely a neural histoid (Gupta et al 2017, Ramanujam et al 1984). High resolution ultrasound is useful in assessment and confirmation.

This is a very interesting case as cell mediated-immune response in Histoid Hansens is very rare. Published literature shows once an abscess is formed with caseous necrosis within the nerve sheath, it does not resolve by medication. Resolution of such swellings or 'abscesses' after 4 weeks of steroid therapy may be interpreted more in favor of inflammation rather than abscesses. Repeat the HRUS after treatment and a wedge biopsy of one of the involved nodular thickening on the nerves could be the approach to prove the point.

The management of nerve abscess depends on the state of nerve function. If there is no loss of function, like our patient, medical management with oral corticosteroids is preferred. Deterioration in nerve function warrants surgical intervention like epineurotomy, aspiration, decompression of the nerve.

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