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Case Report

An intriguing instance of type 2 lepra reaction-induced granulomatous rhinosinusitis: a case report

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ABSTRACT

Hansen disease, or leprosy, is a granulomatous disease caused by the bacteria *Mycobacterium leprae*, which is endemic worldwide. The upper respiratory tract is the main portal of entry for bacilli, and the paranasal sinuses serve as a reservoir of infection and may exhibit features of rhinosinusitis. We presented a man who manifested features of acute rhinosinusitis not respond to standard therapies incidentally found to have type II lepra reaction and showed miraculous recovery of symptoms following oral steroids and patient on close follow-up. Although leprosy has been declared eliminated globally in most countries, otorhinolaryngologists should still consider type II lepra reactions a differential diagnosis whenever a patient presents with nasal symptoms that do not respond to standard therapies.

Keywords: Leprosy, Erythema nodosum leprosum, Rhinosinusitis

INTRODUCTION

Leprosy, is a chronic granulomatous disease caused by the obligate intracellular bacillus, *Mycobacterium leprae* with a worldwide prevalence of 165,459 cases.[1] The detection rate of new cases in India is 73.26 per 1 lakh population and the prevalence in Puducherry is estimated to be 0.1 per 10,000 population.[2] It affects the skin, and peripheral nerves and involves multiple systems, the respiratory tract is the most common transmission route, compassing the nasal septum, turbinates, anterior nasal spine, and paranasal sinus, which serve as an entry point and reservoir for infection.[3,4]

Clinicopathologic manifestations of leprosy are classified into subtypes based on the immune response which is often complicated by immune-mediated responses to *Mycobacterial Leprae* antigen, these lesions present as sudden inflammatory episodes known as Lepra reactions, which require treatment with immunomodulatory drugs.[5]

Erythematous nodosum leprosum (ENL) is a type of lepra reaction commonly involving the skin and other organ systems, but nasal involvement is only observed in 8% of patients. It causes nasal mucosal edema, mucopurulent discharge, nasal crusting, atrophy of turbinates, and rarely septal perforation.[6] Therefore, prompt intervention can avert nasal deformities.

We report an unusual case of type II lepra reaction masquerading as acute rhinosinusitis and a literature review of the same.

CASE REPORT

A 47-year-old man presented with complaints of bilateral progressive nasal obstruction, non-foul smelling mucopurulent nasal discharge, and dysosmia for 2 weeks. He also had dull aching pain in the eyebrows and multiple episodes of anterior epistaxis bilaterally for the past 5 days. No preceding history of nasal trauma, bleeding diathesis, intranasal substance abuse, or nasal surgery was noted.

On examination, the nasal mucosa was diffusely edematous bilaterally, with a pink congested granular

lesion over the cartilaginous part of the nasal septum on the left side covered with mucopurulent discharge (Figure 1) and tenderness over the left frontal, ethmoid and maxillary sinus.

Hematological investigations were within normal range and paranasal sinus X-ray showed a homogenous opacification of the left maxillary and frontal sinus.

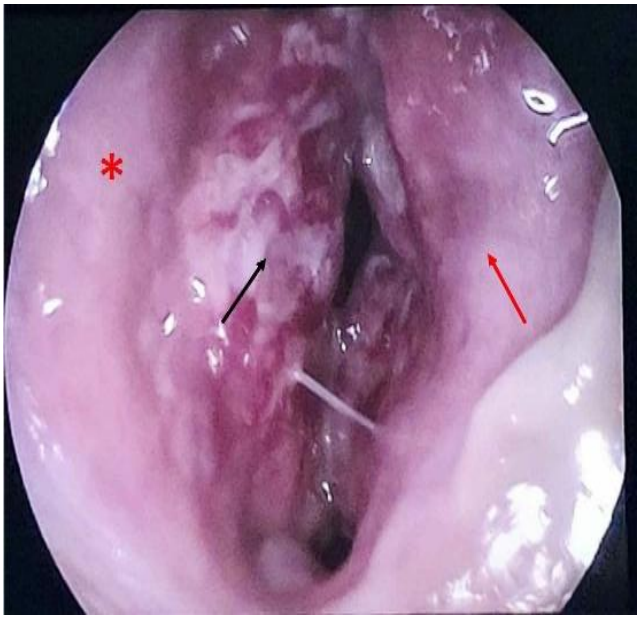

Figure 1: Anterior rhinoscopy shows granular lesions over nasal septum; red asterixis represents oedematous septum, black arrow represents oedematous, infiltrative granular lesions over septum covered with mucopurulent discharge, red arrow represents oedematous and congested lateral wall.

The patient was treated with Intravenous antibiotics (cefotaxime and metronidazole), topical nasal decongestants (xylometazoline 0.05%), mucolytics (bromhexine), and analgesics with a provisional diagnosis of acute rhinosinusitis.

During treatment, the patient was found to have new multiple, well-defined erythematous tender, firm infiltrated nodules of round to oval shape of varying sizes in forearm, trunk, and back (Figure 2). Puzzled by unusual lesions, his history was scrutinized thoroughly and then he disclosed that he had been diagnosed with leprosy 3 years previously and was treated with multidrug therapy (MDT) at government health centre for 1 year and was declared cured.

A dermatologist's opinion was obtained for skin manifestations, and a split skin tissue biopsy was performed on the patient's forearm. Histopathological examination revealed peri-adnexal and perivascular lymphohistiocytic aggregates and the patient was diagnosed with the type II lepra reaction (ENL) (Figure 3).

Figure 2 (A and B): Multiple, erythematous, tender infiltrated nodules of various sizes from 0.5×0.5 cm to 3×3 cm seen in the forearm, trunk, and back (red arrow).

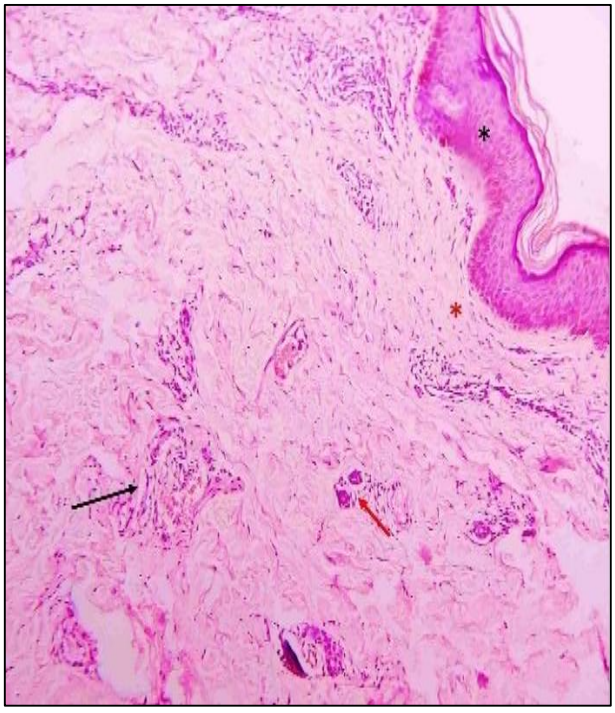

Figure 3: Wedge biopsy of the skin nodule of right forearm showing perivascular and peri adnexal lymphohistiocytic aggregates (H and E, 10×); black asterixis represents atrophic epidermis, red asterixis represents dermis, black arrow shows peri-adnexal lymphohistiocytic aggregates and the red arrow shows perivascular lymphohistiocytic aggregates.

Non-contrast computerized tomography of the nose and paranasal sinus was performed to assess the extent and nature of nasal involvement. The findings revealed a homogenous soft tissue density (mucosal thickening) in the left maxillary and frontal sinus and, focal areas of thickening and thinning of the cartilaginous septum with micro perforation at the posterior end (Figure 4).

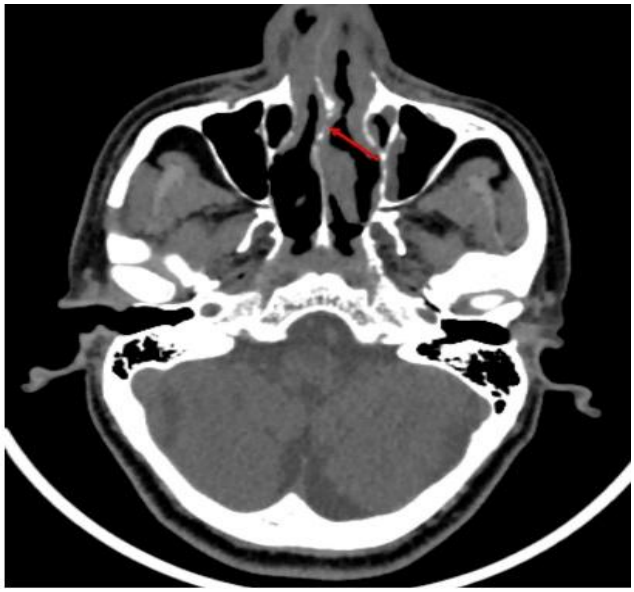

Figure 4: Computed tomography of nose and paranasal sinus axial view showing focal areas of septal thinning and micro-perforation (red arrow).

Since the patient failed to show discernible improvement in symptoms, based on clinical evidence, the patient was started on oral prednisolone at 1 mg/Kg/day for 5 days and then tapered over 1 week. Nasal endoscopic examination performed after 1 week revealed complete resolution of the granular mucosal lesions over the cartilaginous septum (Figure 5).

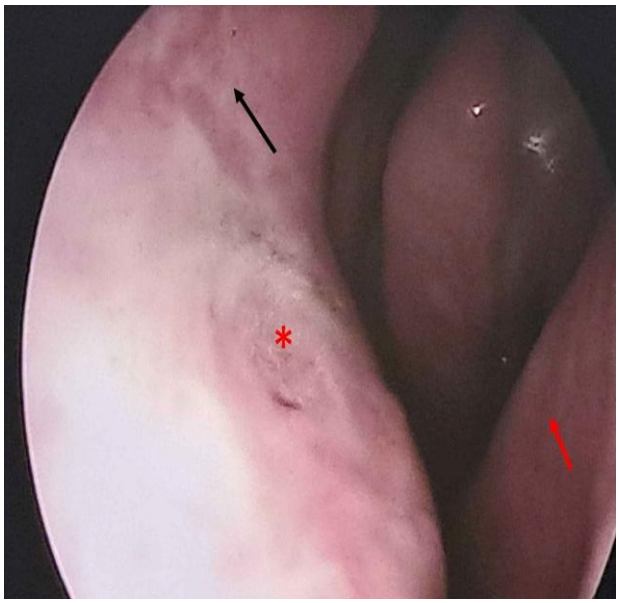

Figure 5: Diagnostic nasal endoscopy showing complete resolution of granular lesions over septum-red asterixis, black arrow shows resolved edema of the nasal septum, and the red arrow shows resolved congestion and edema of the lateral wall.

The skin lesions also exhibited a fair resolution. However, he was again treated with low-dose oral steroids along with 100 mg of oral clofazimine for 1 week to resolve the nodules completely. He has been on regular follow-up for the past 6 months, and there has been no recurrence of symptoms.

DISCUSSION

Leprosy is an infectious disease with high stigmatization that is complicated by acute inflammatory episodes of immunological responses to *Mycobacterium*. Leprae, known as Leprosy reactions, occur before, during, or after the successful completion of MDT. Two distinctive types of lepra reactions have been identified: the type I lepra reaction (reversal reaction) and the type II lepra reaction or ENL.

Type II lepra reactions or ENL, first recognized by Murata in 1912, is a type III hypersensitivity reaction causing deposition of immune complexes leading to the release of proinflammatory cytokines, and neutrophil infiltration results in vasculitis and leads to clinical manifestations in various organs.[5] It is characterized by fever and malaise with crops of tender, erythematous papulo-nodular lesions that affect various organ systems in the body.[7] Histological features include peri-adnexal inflammatory infiltrates (95%), followed by neutrophil aggregates in granuloma (86.9%) due to immune complex deposition.[8]

Vasculitis resulting from ENL can lead to submucosal granular infiltration of the nasal mucosa in the early stages, presenting as nasal obstruction and blood-stained nasal discharge. In the later stages, it can lead to ulceration and focal granuloma formation. Septal perforations have been observed in 8% of patients, and persistent crusting and reduced blood supply to the septal perichondrium may lead to saddle nose deformity if left untreated.[9,10]

Martin et al hypothesized that ENL enabled lepra bacilli to remain viable persistently in the nasal mucosa even after MDT because vasculitis causes reduced blood flow. Their reactivation may lead to hypersensitivity reactions from time to time.[11]

ENL has a variable occurrence before, during, or after completion of therapy (MDT). Pocaterra et al classified ENL based on the number of episodes as Acute single ENL (lasting less than 6 months), acute multiple ENL (more than 1 acute episode lasting less than 6 months), and Chronic ENL (multiple acute episodes lasting more than 6 months).[12] We observed the first variant in our patient.

Although both leprosy and lepra reactions can affect the paranasal sinuses, the propensity of the sinus to be involved varies. ENL commonly affects the ethmoid sinus (80%) followed by the maxillary sinus (48%), whereas leprosy commonly affects the maxillary antrum due to lepromatous infiltration.[13]

Barton et al. described the CT features of the paranasal sinus in lepra reactions as a homogenous soft-tissue density with generalized mucosal thickening.[14] This classical feature correlates well with the observations in this case report.

Corticosteroids are considered the first-line treatment for ENL because they decrease the release of proinflammatory cytokines, and prostaglandin synthetase, which is conducive for neutrophil infiltration thereby suppressing cell-mediated immunity.[15] The administration of oral and parenteral corticosteroids at doses of 1-2 mg per kg body weight and rapid dose tapering within 2-3 weeks is admissible for the relief of lepra reaction episodes.[9]

In pursuance of the abovementioned discussion, treatment with oral steroids alleviated the nasal symptoms in our patient and further continuation of treatment by our dermatology team provided complete resolution of his illness.

CONCLUSION

Type II lepra reaction causes nasal manifestations, such as sequelae of leprosy, even after successful treatment completion. So, whenever a patient presents with nasal manifestations associated with a dermatological condition, an ENT surgeon should consider type II lepra reaction as a differential diagnosis. Hence, profound knowledge about this condition will help ENT surgeons better treat this disease.

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