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Case Report

Corneal Leproma : A Rare Ocular Manifestation in Leprosy

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A leproma is a discrete granulomatous nodule rich in lepra bacilli and is a rare yet characteristic finding seen in the lepromatous spectrum of Hansen's disease. Majority of the ocular lepromas reported in the literature so far arise from uveal tissue and sclera with an incidence of 0.75 to 1%. Exclusive corneal leproma associated with multibacillary leprosy is an extremely rare finding. It is painless and silent nature in earlier stages because of that it may remain unattended for several years until significant visual loss, disease progression and transmission may continue to occur. We report a case having corneal leproma, the detection of which led to the uncovering of advanced cutaneous involvement. The patient had chosen not to disclose his skin involvement and disability possibly due to social stigma and partly due to ignorance. The presence of such untreated multi-bacillary forms in the community may present a significant hurdle for a country like ours, aiming to eliminate this chronic disease. Public awareness through health education coupled with early detection remains the cornerstone in tackling this multi-system disorder which has implications both from disability and transmission angles.

Keywords : Lepromatous Leprosy, Visual Impairment, Corneal Leproma, Histoid Leprosy

Introduction

Hansen's disease, a chronic granulomatous infectious disorder, is more commonly perceived as primarily involving the skin and nerves. However, owing to its multi-system nature, it is possible for this disease to present with extra cutaneous manifestations as well. We report one such highly infectious, hidden case which was brought to the notice of the health care system due to the ocular lesions rather than skin involvement.

Leproma, a granulomatous nodule rich in lepra bacilli is a characteristic feature of Histoid variant. Histoid leprosy, initially described by Wade (1963) is considered to be associated with highly active and resistant infections with a greater risk of widespread transmission.

Lepromas are usually found to be associated with uveal tract and sclera. Since ocular involvement in leprosy occurs due to the spread of lepra bacilli via hematogenous and neural routes, the more vascularised tissues tend to be more commonly involved (Sekhar et al 1994). Isolated corneal involvement is an extremely rare finding. The relatively silent and painless nature of exclusive corneal involvement in the initial presenting stages leads to a significant delay in patients approaching health services for treatment. In contrast, the more commoner uveal tract lepromas will invariably be associated with

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painful, distressing symptoms of accompanying uveitis which prompts earlier presentation and needful interventions.

Case Report

A 42 year old male working in the tourism sector and in constant contact with people from all over the country, was initially seen by a private Opthalmologist from Goa, referred to the Department of Dermatology, Veneorology and Leprosy by SankaraEye Hospital Bengaluru highlighting skin lesions in the patient.

On detailed history, patient revealed that he noticed his ocular morbidity almost 2 years back in the form of gradual painless progressive visual impairment in the left eye associated with noticing a growth over the cornea. Over a span of a few months, it progressively worsened with a further decline in visual acuity, increase in size of corneal mass, redness, photophobia and discomfort (Figs. 1 & 2).

Along with ocular involvement, the patient noticed xerosis, swelling of feet, painful paraesthesias and spontaneous ulcers over hands and feet. The cutaneous involvement gradually progressed to involve the entire body,

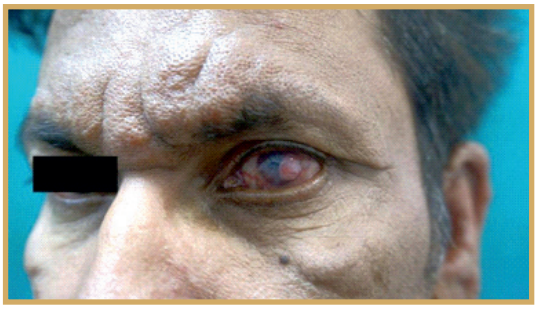

Fig. 1 : Left temporal cornea showing well circumscribed nodular fleshy mass with surface vascularisation. Lateral madarosis and erythematous plaques over the forehead seen.

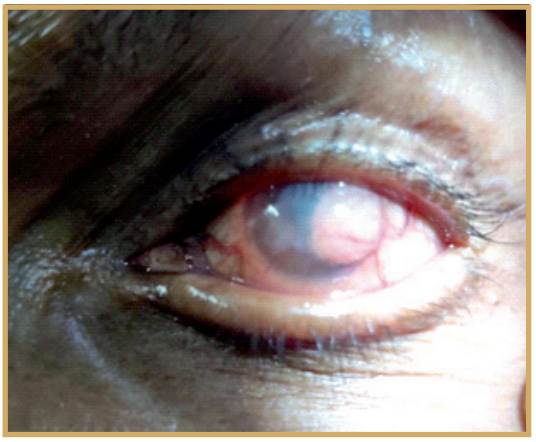

Fig. 2 : Torch light examination of left eye showing well circumscribed corneal mass extending from temporal limbus to mid cornea.

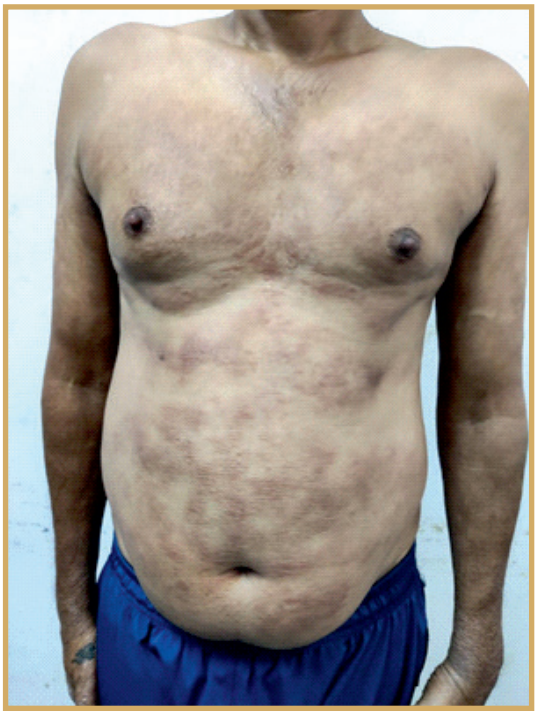

Fig. 3 : Erythematous plaques over abdomen. Gynecomastia was also noted.

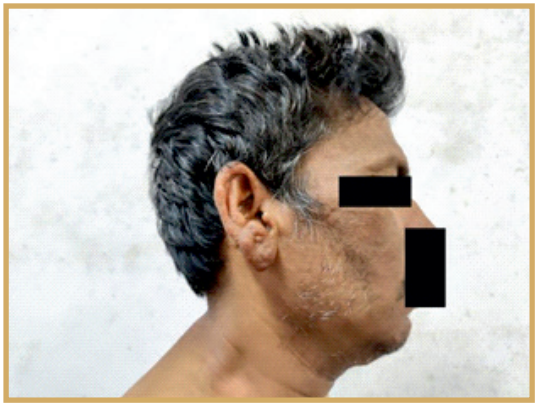

Fig. 4 : Pinna showing presence of nodules.

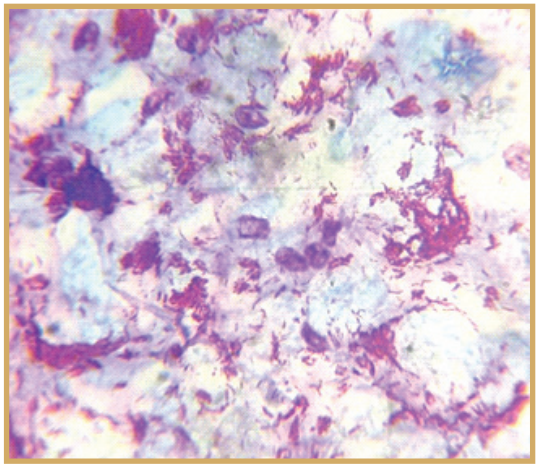

Fig. 5 : Slit skin smear showing clumps of solid staining bacilli, BI- 6+.

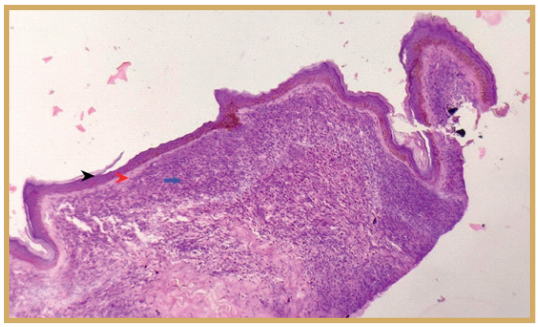

Fig. 6 : HPE (10X) Showing atrophic epidermis (black arrowhead), Clear sub epidermal grenz zone (red arrowhead) with dermis showing dense inflammatory cell infiltrate (blue arrow), few foam cells seen interspersed within dermis.

including trunk, ear lobes and face in the form of erythematous plaques and nodules (Figs. 3 & 4).

Patient received treatment from a local village quack, the course of which lasted for 6 months, after which he discontinued and gives history of significant relief from skin lesions. On eliciting family history patient reluctantly disclosed that his father had similar lesions with associated deformities of hands and feet, causing paralysis and untimely death, the cause of which was unknown. He received treatment with the same village quack. He demonstrated a sense of shame associated with his fathers illness and feared social discrimination and community ostracism.

The patient was subjected to a detailed examination for Hansen's disease, which resulted in findings of thickened nerves, sensory disturbances, and motor weakness with corneal anesthesia and loss of corneal reflex in the involved eye. Lateral madarosis and lagophthalmos were also noted. A bacteriological index of 6+ on Ridley Scale was demonstrated (Fig. 5) and skin biopsy was diagnostic for lepromatous leprosy (Figs. 6 & 7).

Ocular examination by ophthalmologist revealed a well-circumscribed nodular fleshy mass, approximately 6x5 mm in size, encroaching over the corneal surface and limbus with adjoining corneal opacification, circumcorneal congestion and papillary reaction. Slit-lamp biomicroscopy showed significant superficial and deep vascularisation of cornea with dilated tortuous blood vessels. Visual acuity was 6/6

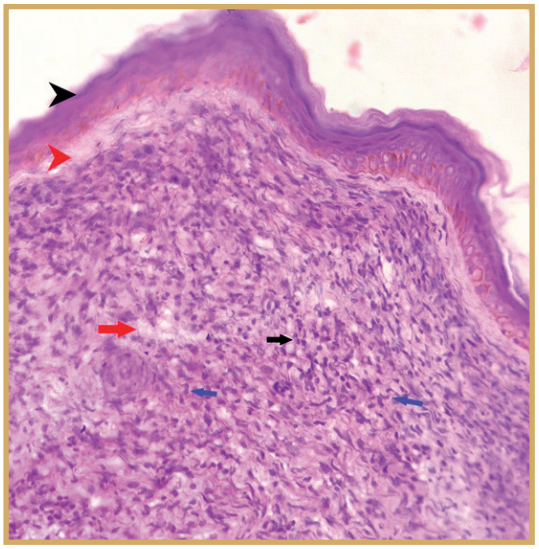

Fig. 7 : Atrophic epidermis, Black arrowhead, (40X HPE); Clear sub-epidermal grenz zone (Red arrowhead); Collection of foamy macrophages (Red arrow); Lymphocytic infiltrate in dermis (Black arrow); Epithelioid cells (Blue arrow).

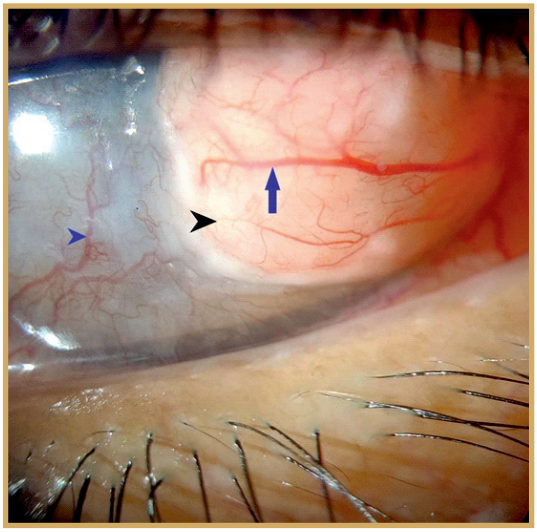

Fig. 8 : Slit lamp bio-microscopy showing well demarcated solitary, fleshy, opaque corneal mass (Black arrowhead) with neovascularisation and opacification of adjoining corneal surface (Blue arrowhead). Tortuous blood vessels seen over mass (Blue arrow).

in the right eye and finger counting at 1 meter in the left eye in case 1. Posterior segment was unremarkable, and intraocular pressure was within normal limits. Excision biopsy of mass and corneal scrapping for AFB was undertaken, which was diagnostic of corneal leproma (Fig. 8).

The patient was subjected to an integrated interdepartmental management. The corneal mass was completely excised and subjected to a histopathological examination. Post excision of leproma patient was started on antibiotic and lubricant eye drops. Multidrug therapy was started after required blood investigations and he was followed up regularly on a monthly basis for one year. There was a significant improvement in skin lesions however, ocular disability, and sensory disturbances continued to remain same.

Discussion

Hansen's is a chronic, multi-system disease with a variable course. Although eliminated from India in 2005, it continues to largely remain endemic, with India contributing to almost 60% of global cases (Rao & Suneetha 2018). India currently ranks among top 3 countries which collectively contribute to 80% of newly detected cases globally (Blok et al 2015). Since the disease has a lengthy, unpredictable course associated with disabilities and social stigma, early detection and effective treatment remain the cornerstone in managing this chronic disorder. Though ocular involvement in Hansen's is known to occur in up to 85% of cases (Reddy & Raju 2006), exclusive corneal lepromas have been very rarely reported, especially in the Indian context.

Ocular associations are worrisome as even after the completion of adequate doses of MB-MDT and resolution of skin lesions, the subsequent development of disabling ocular complications may not be prevented and can also be sight threatening (Grzybowski et al 2015). The presence of ocular involvement generally depends on duration, type of leprosy, number of reactionary episodes and treatment status of patient. The expertise of the medical personnel is also vital in picking up ocular findings in such patients. The most common cause of leprosy related blindness includes corneal disorders like exposure keratitis secondary to lagophthalmos and corneal leucoma (Radhakrishnan & Albert 1980).

Leproma is a superficial, well circumscribed and discrete granulomatous nodule rich in lepra bacilli. Its Incidence is estimated to be around 0.75 to 1 % among lepromatous leprosy patients (Francis et al 2011). It has been reported to be arising in uveal tract, like the highly vascularised ciliary body (Nirankari & Chaddah 1953). Other infrequent locations include limbus (Ram et al 2018), anterior angle (Soni et al 2020), cornea (Yuen et al 2006), and also nasal cavity (Kim et al 2015).

This patient presented to us with an exclusive corneal involvement that was ignored and left unattended in the initial stages of development due to the lack of pain and other distressing symptoms. This led to years of silent progression, which caused disease advancement in the patient. Medical intervention was sought only after significant visual impairment and secondary involvement of nearby structures had already occurred. In contrast to corneal lepromas, the more commoner uveal tract lepromas have a better likelihood of earlier presentation to health care facilities and timely detection due to the painful uveitis and acute, distressing symptoms associated with it.

Since patients presenting with such lepromas usually have multi-bacillary forms of the disease, early identification is of vital importance to prevent community transmission. In addition, such forms are prone to reactionary episodes, which can have devastating effects on the eye as this corneal lepromas can be a part of the reactional episode which makes them prone to globe perforation and consequent permanent vision loss (Rathinam et al 2020).

Corneal lepromas have been very rarely documented in literature so far (Yuen at al 2006) and can often be mistaken for benign neoplastic growths or degenerative conditions. Their presentation in earlier stages give an appearance of a non infectious etiology and leprosy as a causative factor is often overlooked. By presenting these cases we would like to create awareness about this rare manifestation so as to enable prompt suspicion with regard to such lesions that can facilitate earlier intervention and diagnosis.

Our patient had skin, nerve and disabilities along with corneal involvement, which is in tandem to findings reported by Yuen et al (2006). His patient had trophic changes with thickened nerves, whereas in our patient significant skin involvement was also present in addition to nerve, sensory and motor involvement. Our case was perhaps not properly treated/untreated leading to disease progression, presenting as severe, extensive cutaneous involvement. By the time patient presented to us, he was in an advanced form of the disease.

The bacilli in ocular lesions may remain viable even after completion of the full course of treatment and may cause relapses and recurrence

of ocular symptoms. Hence it is vital that such lepromas are picked up at the earliest when the patients present to primary health care facilities. Although regular ocular examination can prove to be very helpful in preventing disabling ocular complications among Hansen's cases (Ffytche 1998), as well as uncovering hidden cases with limited cutaneous involvement or those patients hesitant to come forward, it is practically difficult to conduct such a screening evaluation in the Indian set up due to constrained resources.

Despite having significant skin involvement, our patient did not disclose this most probably due to the stigma associated with leprosy or may be due to lack of knowledge of the multi-system nature of this disease and failure to associate both the conditions to a common causative factor. The ocular findings and subsequent referral to us proved to be the only way by which this patient could be identified and treated which was definitely associated with an unacceptable time lag.

Conclusion

From a public health point of view, such cases form a reservoir of infection in the community causing unabated transmission, which will have a negative impact in our fight against leprosy and is not good for an endemic country. A high index of suspicion is required by general physcians and specialities like Opthalmology for diagnosing such cases early so that transmission is interrupted faster and disabilities are prevented. All clinical disciplines need to be oriented as leprosy can present to various specialities owing to its multi-system involvement. More emphasis on leprosy training in medical curriculum coupled with increasing public awareness to reduce social stigma and to encourage early presentation to health care facilities will go a long way in reducing the burden of this chronic disease.

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