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Case Report

An unusual ocular presentation of histoid leprosy

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ABSTRACT

Leprosy is a chronic granulomatous infection caused by mycobacterium leprae, mainly affecting the skin and peripheral nerve. Histoid type of leprosy, which is considered as an extremely rare variant of lepromatous leprosy. We report a case of a 50/year old male presented to our institute with chief complain of conjunctival swelling in both eyes, encroaching over cornea and involving limbus and sclera, associated with painless blurring of vision. An incisional biopsy of the right eye was performed under local anesthesia. Histopathology of right eye biopsy revealed spindle shaped cells having bland looking nuclei and variable amount of cytoplasm which was suggestive of histoid leprosy. A dermatology consultation was sought and patient was diagnosed to have leprosy. Consequent to which he was started on MBALD (multibacillary anti leprosy drug), and showed improvement in patient`s visual acuity as well as regression of right eye swelling and disappearance of left eye swelling.

Keywords: Conjunctival swelling, Histopathological examination, Histoid leprosy

INTRODUCTION

Leprosy also known as Hansen's disease (HD) is a chronic granulomatous infection caused by mycobacterium leprae. Infection can lead to damage of the nerves, respiratory track, skin, and eyes.[1] Classification of disease based on bacteriological criteria which distinguishing between paucibacillary and multibacillary. Paucibacillary includes tuberculoid and borderline tubercular. Multibacillary leprosy includes remaining borderline form and lepromatous leprosy.[2] There is also a histoid type, which is considered as an extremely rare and uncommon variant of lepromatous leprosy which included in multibacillary infection.[2] Ocular manifestations include madarosis, cutaneous nodules on the lid, lagophthalmos secondary to paralysis of orbicularis oculi. In cornea earliest sign is beaded corneal nerves, superficial and interstitial keratitis, pannus are late signs, dry eye in lepromatous sepctrum, episcleral nodules are small chronic nodules, commonly situated at limbus and sometimes extend into the cornea and it is cardinal sign of deep conjunctival tissue involvement, and uveitis quite common and most frequent manifestation in leprosy. Episcleritis is more common than scleritis. Iris pearls are small, round, white and shiny lepromata near pupillary margin are pathognomonic of leprosy, Conjunctival nodules, scleritis and lesion in retina and posterior pole involvement are rare.[1,2] Here, we describe a patient of histoid leprosy who presented only with ocular manifestations and no other systemic involvement.

CASE REPORT

A 50 year/old male, farmer by occupation presented to us with the complaint of swelling over the conjunctiva and decrease of vision in both eyes since3 months. The swelling was gradually increasing in size and encroaching cornea, associated with painless blurring of vision. It was not associated with pain, discharge, watering, redness or photophobia. There was no previous history of trauma or surgery of eye. Family, personal or social history was

insignificant. On ocular examination, visual acuity in Right eye was 20/320 and left eye was 20/60. Loss of eyebrows was seen in both eyes. There was mild ectropion and tylosis in both eyes, rest ocular adnexa were normal. On slit lamp examination, right eye swelling was 8×6×2mm in size extending from 3to6`o clock and left eye swelling was 4×2×1 mm in size extending from 6to8`o clock involving cornea, limbus and sclera (Figure 1).

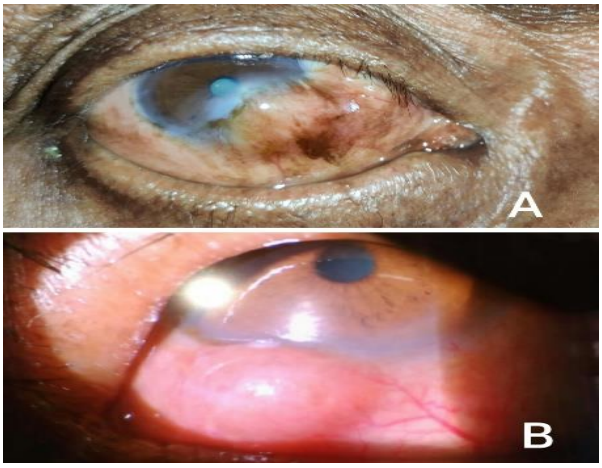

Figure 1: (A) Right eye which shows 8×6×2mm sized conjunctival swelling involving cornea, limbus and sclera (B) initial presentation of left eye conjunctival swelling which is smaller than right eye 4×2×1mm in sized.

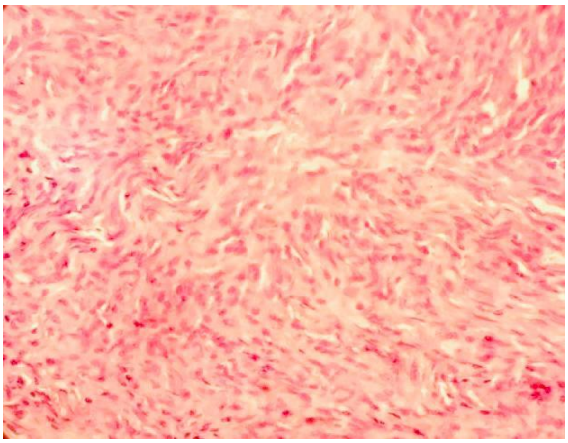

Figure 2: The histological findings show spindle shaped cells, having bland looking nuclei and variable amount of cytoplasm suggestive of histoid leprosy.

There was interstitial keratitis in inferonasal in right eye which was extending up to mid pupillary area from 3 to7`o clock with superficial vascularization of cornea and mild inferotemporal keratitis in left eye. Corneal sensations in both eyes were decreased. Both eyes showed cataractous changes, rest anterior segment was within normal limits. Intraocular pressure was 17.3mmhg in both eyes. Dilated fundus examination was essentially normal.

On general examination, the patient was moderately built, well- nourished, oriented to time place and person. On examination multiple shiny skin coloured to dull brown nodular lesions over the back and face. No other hypo/ hyperpigmented lesions elsewhere on the body. An incisional biopsy of right eye conjunctival-limbal swelling was performed under local anesthesia and sent in formalin container for histopathological examination. Histopathological study revealed spindle shaped cell having bland looking nuclei and variable amount of cytoplasm suggestive of histoid leprosy and fite faraco satin positive (Figure 2). Patient was started on moxifloxacin 0.5% topical antibiotic eye drops 4 times a day, fluorometholone0.1% eye drops 4hourly in both eyes and chloramphenicol and polymyxin-B-Sulphate ophthalmic ointment at night.

The patient was referred to the dermatologist for further management of leprosy where further examination revealed bilateral non tender uniformly thickened ulnar, superficial radial, greater auricular and right sided common peroneal nerve thickening. There was loss of temperature differentiation ability on bilateral hands and feet in glove and stocking distribution, with the other sensations and motor functions intact. On our clinical n histopathological findings, we were confirmed the diagnosis of leprosy and he was started WHO recommended multibacillary anti leprosy drug (MBALD) regimen. On follow up at 6 weeks, the patient was symptomatically better. On ocular examination, visual acuity (UCVA) improved to (R/E) 20/80, (L/E) 20/40 pinhole visual acuity in (R/E)20/40, (L/E) 20/30. There was minimal perilimbal congestion and conjunctival swelling had significantly reduced in size and extent in right eye and disappeared in left eye (Figure 3).

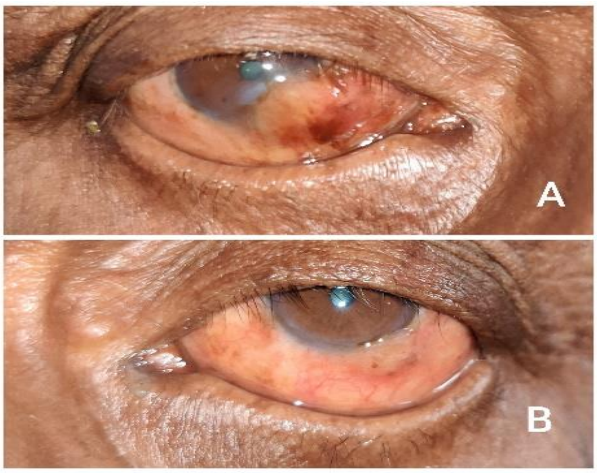

Figure 3: (A) right eye shows regression in conjunctival swelling after 6 weeks of multi bacillary anti leprosy drug regimen. (B) left eye conjunctival swelling disappears after 6 weeks of multi bacillary anti leprosy drug regimen.

Interstitial keratitis regressed from the pupillary area to beneath the pupillary border from 4 to 6`o clock. The

frequency of fluoromethalone 0.1% eye drops was reduced four times a day for two weeks; then tapered to stop over a month time. His systemic medication regimen is to be continued for further period of 12 months and the patient was reviewed conjointly in both ophthalmology and dermatology clinic monthly.

DISCUSSION

Leprosy is responsible for 5% of blindness worldwide.[5] It has the highest incidence of ocular involvement than any other single systemic disease.[7] Ocular involvements is reported to be as high as 85.5%, with the highest involvement in lepromatous leprosy.[8,9] The incidence of leproma of the eye is reported to be 0.75 to 1% among lepromatous leprosy patients. The ocular involvement in leprosy may be due to (a) primary infection of the ocular adenexa; (b) secondary to nerve involvement (c) due to direct invasion of the anterior segment of the eye with gradual spread to conjunctiva and cornea; or (d) sensitization of ocular tissues by the presence of mycobacteria either locally or elsewhere in the body. Primary ocular manifestations of leprosy include madarosis, lagophthalmos and ectropion of the lids, chronic conjunctivitis, corneal nerve thickening, interstitial keratitis, corneal ulcer, episcleritis and/or scleritis and uveitis. In general, the ocular clinical picture of leprosy is usually mixed, with multiple features involving several eye structures. In this case, the ocular presentation was confined to the conjunctiva, limbus and cornea only, with no other ocular manifestations. Histoid leprosy is a well-recognized rare expression of multi-bacillary leprosy with characteristic clinical and histopathological features.[10-15]

This condition was first described by Wade in 1963.[5] The histoid lesions commonly appear as smooth, hemispherical, non-tender, soft to firm subcutaneous nodules on an otherwise normal looking skin. Histologically, these lesions are composed of abundance of histiocytes with copious amounts of acid-fast bacilli. The prevalence of the histoid variant of lepromatous leprosy is about 1.8% to 3.6% of all lepromatous leprosy cases.[16,17] It is mostly reported in patients who had received dapsone as monotherapy for the treatment of their leprosy. Some cases were reported several years after effective completion of multi-drug therapy, as a relapse.[15] Though extremely rare, it has also been reported in newly diagnosed patients who have not received any treatment.[16]

Our patient developed histoid variant of lepromatous leprosy de novo, as he never had history of leprosy or contact with known leprosy patient. Conjunctival-limbal swelling or limbal leproma were typically seen on both sides more so on an both eye near limbus the leproma were non tender due to impairment of sensations. Biopsy of one of these leproma confirmed the etiology. The lenticular changes in both eyes appeared to be senile. The presence of interstitial keratitis, leproma, madarosis was a very distinctive feature which has been described to be characteristic of leprosy. The local and general treatment as described above was carried out for 2 and half month. Limbal leproma was regressed in right eye and disappeared in left eye with in 2 and half month. The general treatment of MBALD regimen was continued for 12 months. This is a rare case of histoid leprosy with solitary eye involvement without any other systemic signs and skin lesions.

CONCLUSION

Histoid leprosy which is a rare variant of lepromatous leprosy which can present solitarily with ocular manifestations like conjunctival-limbal-scleral swelling, interstitial keratitis and loss of eyebrows and eyelashes without any other systemic involvement. Thus, thorough examination, appropriate intervention and histopathological assessment can lead to diagnose and start treatment of patient early with MBALD regimen. A high index of suspicion for leprosy should kept in mind for patient presenting with conjunctival-limbal nodule or episcleral nodule. Moreover, leprosy should be an essential differential diagnosis of conjunctival- limbal-scleral swellings and interstitial keratitis.

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