

Case report

Conjunctival rhinosporidiosis presenting as a cystic mass-an unusual presentation

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Abstract

Rhinosporidiosis is endemic in India and Sri Lanka and parts of East Africa and South America. Cases of ocular rhinosporidiosis have also been reported from Nepal. Ocular rhinosporidiosis usually involves the conjunctiva and the lacrimal sac. It usually presents as a polypoidal mass. Cystic mode of presentation has not yet been reported in the literature. We herein report an unusual mode of presentation of ocular rhinosporidiosis presenting as a huge conjunctival cystic mass.

Key words: conjunctival cyst, oculosporidiosis, rhinosporidiosis, Rhinosporidiosis seeberi

Introduction

Rhinosporidiosis is a chronic infection caused by Rhinosporidiosis seeberi, long considered as a fungus, but now classified under a new class - the Mesomycetozoa (Arseculeratne SN, 2005). It usually produces granulomatous inflammation of the nose and nasopharynx. Ocular rhinosporidiosis usually involves the conjunctiva and the lacrimal sac. The typical ocular manifestations of rhinosporidiosis (Oculosporidiosis) are conjunctival, lacrimal or eyelid infections (Arseculeratne SN, 2002).

Oculosporidiosis clinically appears as a freely mobile, granular, pink, fleshy, sessile or pedunculated mass (Arseculeratne SN, 2002).

Cystic mode of presentation of ocular rhinosporidiosis has not yet been reported in the literature. We are reporting this case of conjunctival rhinosporidiosis having a very unusual clinical presentation in the form of huge conjunctival cyst.

Case report

A 35-year-old farmer presented with complaints of

a mass in his left eye, associated with watering and redness of three months duration. The mass began as small pea size on the conjunctiva which gradually increased to the present size. There was history of swimming in the local ponds. There was no history of trauma, discharge, surgery or chronic exposure to sunlight.

On examination, the right eye was normal. Visual acuity of the left eye was 6/6. Extra-ocular movements of the left eye were restricted in down gaze because of the mass. There was mechanical ectropion of the left lower lid. There was a mass in the left eye involving whole of the inferior bulbar conjunctiva. (Figure 1) The mass was pedunculated and cystic. The surface of the mass was smooth without any bleeding on touch. It felt like a bag of worms. The transillumination test of the mass was positive. Other ocular findings were within normal limits. Ultrasonography A- scan and B-scan showed a homogenous mass with moderate internal reflectivity. Systemic examination revealed no abnormality.



Fig 1: Cystic mass (4×3 cm) involving the whole of the inferior bulbar conjunctiva.

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A complete excision of the cystic mass was done under local anesthesia. Intra-operative findings showed jelly like contents inside the cyst. The conjunctiva was sutured with interrupted 8-0 vicryl suture.

Post-operatively the patient was prescribed topical antibiotics drops with anti-inflammatory drugs systemically. (Figure 2)



Fig 2: Post-operative photograph

Histopathology of the specimen revealed keratinized stratified epithelium with exocytosis. The sub-epithelial area showed oedema with numerous sporangia containing sporangiospores, destruction of sporangium releasing sporangiospores into the stroma, surrounded by inflammatory cells comprised of lymphocytes, histiocytes and neutrophils. Occasional intraepithelial sporangia were also seen. On the basis of these histopathological findings, the diagnosis of rhinosporidiosis of the left inferior bulbar conjunctiva was made. (Figure 3 and 4)

There was no evidence of any recurrence after six months of follow-up.

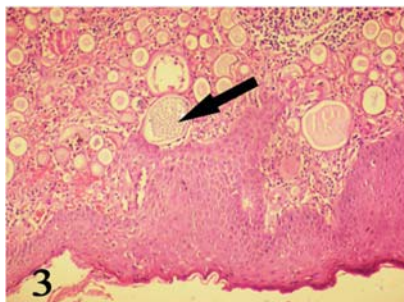


Fig 3: Conjunctival specimen : Keratinized stratified squamous epithelium lining with subepithelium revealing numerous sporangia (arrow head) with surrounding edema and inflammation. (Stain H and E, 100X)

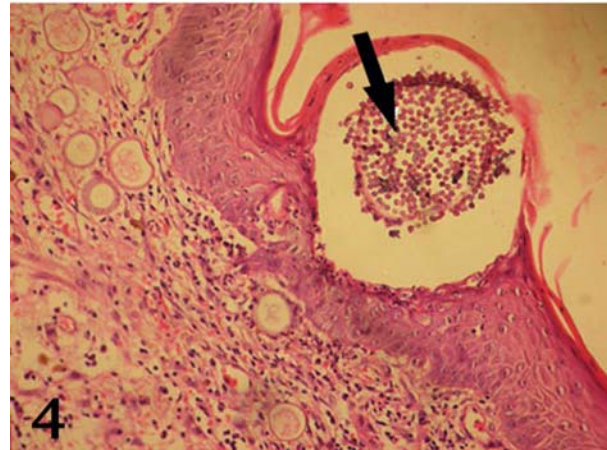


Figure 4: Conjunctiva: Intra-epithelium sporangium (arrow head) with sporangiospores. (Stain H and E, 200X)

Discussion

Rhinosporidiosis is endemic in South Asia notably India and Sri Lanka (Arseculeratne SN, 2002). Although rhinosporidiosis is not endemic in Nepal, isolated cases have been reported from Nepal (Shrestha et al. 1998; Thakur SKD et al. 2002).

Rhinosporidiosis is an infective disease caused by *Rhinosporidium seeberi* where the pathogen is always found in the affected tissue. The majority of cases are sporadic. The presumed mode of transmission of rhinosporidiosis is direct invasion through traumatized epithelium (Arseculeratne SN, 2002).

The majority of cases occur in the upper respiratory sites, notably the anterior nares, nasal cavity, nasopharynx, larynx, soft palate and buccal cavity. The eye and adnexa are the second most common sites of *Rhinosporidium seeberi* infection in human (Arseculeratne SN, 2002).

Kuriakose T (1963) coined the term Oculosporidiosis for ocular rhinosporidiosis. On reviewing the literature, it appears that 4,848 cases have been reported so far out of which 4,040 (83.3%) were nasal, 546 (11.2%) were ocular and 262 (5.4%) involved other sites (Billore, 1996).

Bizarre sites of rhinosporidiosis have also been mentioned in the literature like in skin as subcutaneous nodules, genital organs, trachea and bronchus. About 15 % of cases involve ocular structures, mainly bulbar or palpebral conjunctiva, lacrimal sac, or nasolacrimal duct (Arseculeratne SN, 2002).



In ocular rhinosporidiosis, the palpebral conjunctiva is involved more commonly than the bulbar conjunctiva, caruncle or canthi (Billore OP, 1996). Kuriakose T (1963) has reported 25 cases of ocular rhinosporidiosis out of which 64 % had involvement of palpebral conjunctiva and 24 % lacrimal sac.

In a study from Nepal, Shrestha et al. (1998) reported involvement of the conjunctiva in 76 (92.6 %) and lacrimal sac in 6 (7.3 %) cases. Cases with conjunctival involvement presented as conjunctival polyps. Another study from Nepal, Thakur SKD et. al (2002), reported five cases of oculosporidiosis. Out of these, three cases were seen with involvement of lacrimal sac, one presented as mass in the lower fornix and one with a history of recurrent chalazion.

Shukla et al. (1982) reported 19 cases of oculosporidiosis, of which 71 (59.6 %) involved the palpebral conjunctiva and 38 (31.5 %) involved the lacrimal sac. Palpebral conjunctiva has greater predilection for rhinosporidiosis than any other part of the eye. The exposed bulbar conjunctiva is least affected (Billore OP. 1996).

Scleral melting and staphyloma have been documented secondary to conjunctival involvement, and may be seen as initial findings (Billore OP. 1996). Oculosporidiosis presenting as an under eyelid swelling too has been reported in the literature (Ghorpade A et. al 2007).

On reviewing the literature, it is found that the commonest mode of presentation of conjunctival rhinosporidiosis is a friable, richly vascularized, polypoidal growth that may be pedunculated or sessile with “strawberry” appearance. The surface is covered with tiny white spots consistent with underlying mature sporangia, beneath the epithelium (Arseculeratne SN, 2002).

The gross appearance, though distinctive, is not diagnostic. In this case, conjunctival rhinosporidiosis presented as a huge cystic lesion involving the inferior bulbar conjunctiva looking like a conjunctival inclusion cyst or any parasitic cyst. Histopathological examination of the excised cyst confirmed the diagnosis of rhinosporidiosis. Hence it is clear that definite diagnosis of rhinosporidiosis is possible with biopsy, which will show trophocytes and sporangia in all stages of development (Arseculeratne SN, 2002). Meticulous excision of conjunctival rhinosporidiosis is the treatment of choice, although rare cases of spontaneous regression have been reported (Arseculeratne SN, 2002; Shrestha SP et al. 1998). The potential for recurrence is due to spillage of endospores on adjacent mucosa.

Recurrences are rare (10 %) which can be prevented by cauterization of the base of the lesion or alternatively by cryotherapy (Shrestha SP et al. 1998). Medical therapy is still controversial since cultures of R. Seebri have been unsuccessful in all artificial culture media thus making drug sensitivity impossible. Dapsone (4, 4-diaminodiphenyl sulphone) is the only drug that has some success in treating R. Seebri. It acts by arresting maturation of sporangia and accelerating their degenerative changes (Vijaikumar M et al. 2002). In this case also, complete excision of the cyst showed no recurrence till six months of follow-up.

To conclude, clinical presentation of oculosporidiosis varies. Any huge cystic mass involving the bulbar conjunctiva should arouse the suspicion of ocular rhinosporidiosis. Complete excision with or without cauterization or cryotherapy of the cyst is the treatment of choice.

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