Original Article

Clinical Spectrum and Management Outcome of Ocular and Adnexal Rhinosporidiosis

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Abstract

Purpose: To describe the clinical spectrum and management outcomes of ocular rhinosporidiosis.

Methods: All histopathologically diagnosed cases of ocular rhinosporidiosis between January 2000 and December 2016 were included in the study. The lesions were classified based on the site of involvement, namely conjunctiva, lacrimal sac, eyelid, and orbit. The frequency and percentages for each of the lesions and the different treatment modalities were noted and calculated. Any recurrence and its subsequent management were also noted.

Results: A total of 34 patients were included with a male-to-female ratio of 2.7:1. Conjunctiva was the most common site involved (19, 55.8%), followed by lacrimal sac (11, 32.3%) and eyelid (3, 8.82%). One patient had orbital involvement secondary to sinonasal extension. The mean duration of symptoms was 14.8 ± 19.1 months (range, 1–84 months). Seven (36.8%) patients in the conjunctival group needed scleral patch graft. Five patients (45.4%) with lacrimal sac rhinosporidiosis underwent dacryocystectomy and modified dacryocystorhinostomy (DCR). The mean follow-up period was 5.43 ± 7.9 months (range, 1–36 months). Five (14.7%) patients (2 conjunctival, 2 lacrimal, and 1 eyelid) had recurrence.

Conclusions: Conjunctiva is the most common site for ocular rhinosporidiosis, followed by lacrimal sac. While conjunctival lesions respond well to complete excision with cauterization of the base, lacrimal sac lesions can be managed by a modified DCR. Eyelid rhinosporidiosis can mimic a neoplasm and should be considered in differential diagnosis in suspicious lesions, especially in endemic areas.

Keywords: Conjunctiva, Eyelid, Lacrimal, Orbit, Rhinosporidiosis

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Introduction

Rhinosporidium seeberi was first described by Seeber of Argentina in 1900, and the first independent case was reported by O'Kinealy from Calcutta Medical College, India, in 1903.[1,2] Earlier believed to be a fungus, it was later classified as a *Mesomycetozoa*, the class which includes amphibian protistan pathogens.[3] Rhinosporidiosis is a chronic infection affecting most commonly the mucous membranes of the nose and paranasal sinuses. Ocular involvement has been noted in 15% of the cases.[3] The disease is endemic in India, Sri Lanka, Bangladesh, and Nepal, with pockets of clusters in different states across the country. Although there are studies available on rhinosporidial infection from India, it is surprising that there are very few focusing on the entire spectrum of ocular rhinosporidiosis.[4-7] The studies by Moses *et al*. and Suseela and Subramaniam were conducted 30 and 50 years back and mentioned only the epidemiology of ocular rhinosporidiosis without stating much about the management.[4-7] While Chowdhury *et al*. and Mithal *et al*. have described the clinical spectrum of ocular rhinosporidiosis, but they have not talked

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much about the management and recurrence.[6] Since then, the disease has seen many changes in the management protocols, and there is a need for a study in the literature from India discussing the current scenario of ocular rhinosporidiosis and its management in detail. Concerning these lacunas of literature, the present study was carried out to describe the complete clinical spectrum of ocular rhinosporidiosis and its management outcome at a tertiary eye care center in India.

Methods

It was a retrospective study wherein all patients histopathologically diagnosed with ocular and adnexal rhinosporidiosis between January 2000 and December 2016 were included. Institutional review board approval was obtained, and the study adhered to the tenets of the Declaration of Helsinki. Demographic details, clinical and radiological (if relevant) findings, and management outcomes were analyzed. The lesions were classified based on the site of involvement, namely conjunctiva, lacrimal sac, eyelid, and orbit, and the treatment modalities for each of them were noted. The frequency and percentages for each of the lesions and the different treatment modalities were also noted and calculated. Conjunctival lesions were completely excised with cauterization of the base. Amniotic membrane transplantation and scleral patch graft were done in cases of exposed ocular surface and sclera thinning with or without staphyloma formation, respectively. Most of the lacrimal sac lesions were identified intraoperatively and were managed either by a dacryocystectomy (DCT) or modified dacryocystorhinostomy (DCR). Proper adequate cauterization of the surrounding area and the residual nasolacrimal duct was done in cases of DCT. DCR was performed by the modified DCR technique as proposed by Nuruddin *et al*.[8] Here, the posterior lacrimal sac flap was completely excised, while the anterior flap was trimmed to leave a small stump. Before suturing the flaps, the intrasac granuloma was completely excised, and the area surrounding the common internal ostium and the opened nasal cavity was packed with 5% povidone iodine-soaked gauze for 5 min. Cauterization was avoided to prevent fibrosis. Intubation was performed on surgeon's discretion, and it was removed at 6 weeks. The success of the procedure was defined as significant improvement in watering subjectively and patent lacrimal sac irrigation. Any recurrence and its subsequent management were noted. The patients gave consent for publication of photographs and clinical details for research purpose.

Results

A total of 34 patients were included in the study. A male preponderance was noted with male-to-female ratio being 2.7:1. Conjunctiva was the most common site of involvement (19, 55.8%), followed by lacrimal sac (11, 32.3%) and eyelid (3, 8.82%). One patient had orbital involvement secondary to sinonasal extension. The mean duration of symptoms was 14.8 ± 19.1 months (range, 1–84, months) [Table 1].

Table 1: Demographic and clinical details of the study population

Parameters	*n* (%)
Sample size	34
Mean±SD age (years) (range)	26.68±14.6 (6-60)
Mean±SD duration of symptoms (months) (range)	14.8±19.1 (1-84)
Sex	
Male	25 (73.5)
Female	9 (26.4)
Eye	
Right	17 (50)
Left	17 (50)
Site	
Conjunctiva	19 (55.8)
Lacrimal sac	11 (32.3)
Eyelid	3 (8.82)
Orbit	1 (2.9)
Clinical presentation and features	
Reddish sessile conjunctival mass	19 (55.8)
Swelling in the medial canthal region	10 (29.4)
Scleral thinning	9 (26.4)
Status postlacrimal surgery	3 (8.82)
Eyelid mass	3 (8.82)
Nasal mass	2 (5.8)
Painful proptosis	1 (2.9)
Clinical diagnosis	
Pyogenic granuloma	8 (23.5)
Nasolacrimal duct obstruction	6 (17.64)
Rhinosporidiosis	4 (11.7)
Eyelid tumor	2 (5.8)
Lacrimal sac malignancy	1 (2.9)
Not specified	13 (38.2)
Management	
Excision with cauterization of the base±amniotic membrane transplant/scleral patch graft	15 (44.1)
DCT	5 (14.7)
Modified DCR	5 (14.7)
Mass excision±eyelid reconstruction	3 (8.82)
Medial orbitotomy	1 (3.3)
Adjuvant oral dapsone	2 (5.8)
Recurrence	5 (14.7)
Mean follow-up (months) (range)	5.43±7.9 (1-36)

DCR: Dacryocystorhinostomy, DCT: Dacryocystectomy

In the conjunctival group, 13 (38.2%) patients had involvement of bulbar conjunctiva, out of which 4 (11.7%) had masses near the limbus. Five (14.7%) patients had involvement of the tarsal conjunctiva. One patient had both bulbar and tarsal conjunctival involvement. Patients with isolated tarsal conjunctival involvement presented with reddish pedunculated granular mass, while those with involvement of the bulbar conjunctiva presented as a reddish strawberry-like sessile lesion with characteristic pinhead-sized yellowish dots on the surface in all patients [Figure 1a-c]. Nine (26.4%) of these patients had associated scleral thinning.

Lacrimal sac rhinosporidiosis presented as swelling in the medial canthal region with features of nasolacrimal duct

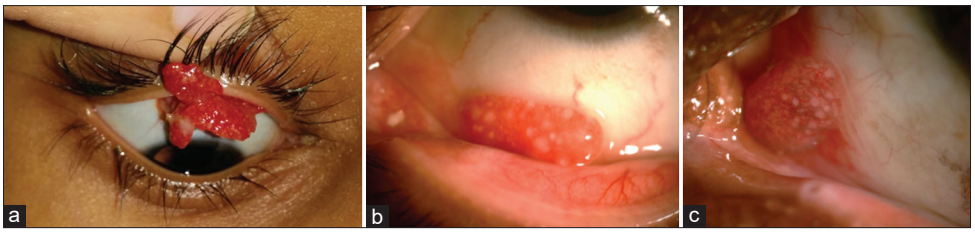

Figure 1: (a) External photograph showing a reddish mulberry-like lesion from the upper palpebral conjunctiva resembling a pyogenic granuloma. (b and c) Slit-lamp photograph showing a reddish mass from the inferior and bulbar conjunctival surfaces respectively. Notice the pinhead-sized yellowish dots over the surface of the lesion depicting rhinosporidial sporangia

obstruction in 10 (90.9%) cases [Figure 2a-d]. Two cases were presented after a DCR, while one after a DCT performed elsewhere for presumed primary acquired nasolacrimal duct obstruction (PANDO). All three had a persistent mass in the lacrimal sac region. All patients with lacrimal sac involvement underwent diagnostic nasal endoscopic (DNE) examination. DNE examination of one of these patients revealed mulberry-like lesions over the inferior turbinate, while another had inferior turbinate inflammation. One patient had an associated nasal mass in the inferior meatus.

Three (8.82%) patients presented with eyelid mass. Two of these had lower lid involvement while one had involvement of the upper lid. The initial diagnosis was rhinosporidiosis in one of the cases, while it was neurofibroma and eyelid lymphoma in the other two. One of the cases had already undergone surgery elsewhere 15 years earlier, but the diagnosis was not available. The eyelid masses were approached via standard lid crease incision, and debulking of the mass was done as much as possible.

One patient had sinonasal mass with orbital extension [Figure 3a and b]. The patient presented with painful proptosis. This patient had undergone some lacrimal sac surgery elsewhere previously. The patient had a mass in the oropharynx too [Figure 3c]. The imaging showed involvement of the medial orbit [Figure 3d]. Incisional biopsy confirmed the diagnosis of rhinosporidiosis, and the patient had marked clinical improvement after debulking of the mass and 6 months of oral dapsone therapy [Figure 3e and f]. The debulking was carried out via an inferior orbitotomy by conjunctival approach, along with the removal of sinonasal mass by an ENT surgeon in the same sitting.

Patients with conjunctival rhinosporidiosis underwent complete excision of the mass with cauterization of the base. Seven (36.8%) patients needed scleral patch graft. Four of these patients underwent staphylectomy, while the other three had an underlying thinning of the sclera. One patient each needed an amniotic membrane transplant and a conjunctival autograft.

Five (45.4%) patients with lacrimal sac rhinosporidiosis underwent DCT, while another 5 (45.4%) underwent modified DCR. One patient with lacrimal sac rhinosporidiosis had undergone endonasal DCR, and a suspicious mass was found intraoperatively which was sent for biopsy. He was later advised DCT but was lost to follow-up. One of the patients

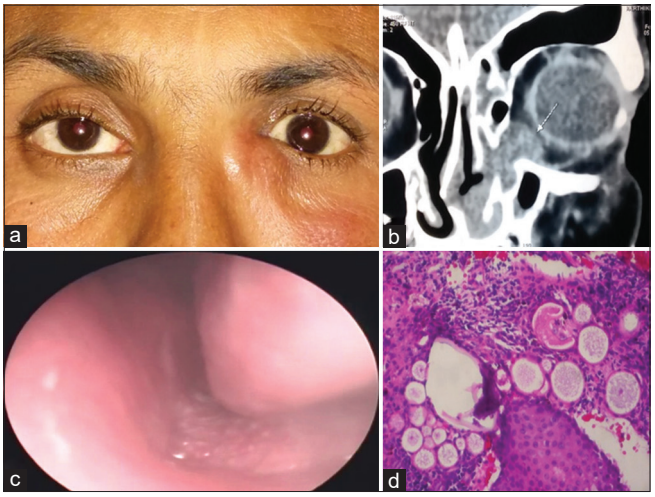

Figure 2: (a) External photograph showing swelling in the left lacrimal sac region. (b) Computed tomography scan of the orbit showing a soft tissue lesion in the left lacrimal sac extending into the inferior meatus. (c) Nasal endoscopy showing mass in the inferior meatus. (d) Microphotograph showing rhinosporidial sporangia in different stages of maturation

who underwent a DCT also needed an inferior turbinectomy and complete removal of the nasal mass by an ENT surgeon in the same sitting. A bicanalicular intubation was done in four out of five (80%) patients who underwent a modified DCR. Four of these patients (80%) had a patent lacrimal system with minimal or no watering at the last follow-up, while one developed a common canalicular block.

One out of the three patients with eyelid rhinosporidiosis underwent debulking of the mass without sacrificing any lid tissue. In this particular case, the preliminary diagnosis was eyelid lymphangioma. Two patients needed complete excision of the mass with eyelid reconstruction by direct closure and Hughes's tarsoconjunctival flap with cheek advancement.

Two patients (one lacrimal and one orbital) received adjuvant dapsone (Dapsone, Glaxosmithkline, 100 mg BD) after ruling out G6PD deficiency for 3 and 6 months, respectively.

A preliminary clinical diagnosis of rhinosporidiosis was considered in only 4 (11.7%) cases, while no specific diagnosis could be made in 13 (38.2%) cases. Eight (42.1%) of the conjunctival cases and 6 (54.5%) of the lacrimal cases were diagnosed with pyogenic granuloma and PANDO, respectively. Two out of three (66.6%) cases of eyelid rhinosporidiosis were

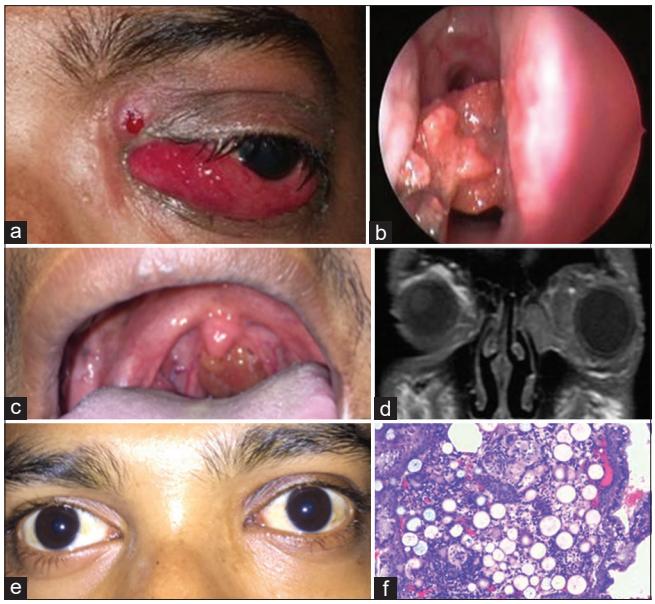

Figure 3: (a) External photograph showing left eye proptosis and chemosis. Notice the scar in the lacrimal sac region from the previous surgery. (b) Nasal endoscopy showing mass in the nasal cavity. (c) Rhinosporidiosis of the oropharynx. (d) Magnetic resonance imaging orbit showing mass in the medial orbit extending to the lacrimal sac region, ethmoid sinus and nose. (e) Resolution of proptosis and chemosis after debulking and dapsone therapy. (f) Microphotograph showing rhinosporidial sporangia in different stages of maturation

clinically diagnosed as benign eyelid tumor (neurofibroma and lymphangioma).

The mean follow-up period was 5.43 ± 7.9 months (range, 1–36 months). One patient had a follow-up of 18 years. A total of 5 (14.7%) patients (2 conjunctival, 2 lacrimal, and 1 eyelid) had recurrence. The conjunctival and lacrimal recurrence was treated with re-excision of the mass. One of these patients with lacrimal recurrence received adjuvant oral dapsone for 3 months. The patient with recurrent eyelid mass underwent wide excision with eyelid reconstruction by Cutler Beard technique.

Discussion

Rhinosporidiosis is a chronic infection commonly affecting the mucosa of the nose and paranasal sinuses. It is caused by *Rhinosporidium seeberi*, an aquatic parasite belonging to the group *Mesomycetozoa*. Ocular rhinosporidiosis accounts for around 15% of all rhinosporidial infections.[3] In the present study comprising 34 cases, we noticed a male preponderance with conjunctiva as the most common ocular site getting affected. Both these findings have been substantiated by previous studies.[7,9] Conjunctival involvement was noted in more than half of our cases (55.8%), and the reported involvement in the literature ranges from 50% to 90%.[7,9] Bulbar conjunctiva is the most common site to be involved because it is the most exposed portion, and we noted a similar finding. The typical conjunctival lesions present as a red, fleshy, polypoidal mass which is sessile when arising from bulbar conjunctiva and sessile or pedunculated when arising from the palpebral or tarsal conjunctiva. Most of the lesions have typical pinhead-sized yellowish dots over the surface of the lesion representing the rhinosporidial sporangia. It is a clinical feature that can make the diagnosis quite easy. Although this typical finding was noted in all of our bulbar lesions, most of them were considered pyogenic granuloma. The chances of the lesion getting misdiagnosed as pyogenic granuloma are more in cases of lesions arising from the palpebral surfaces. There are reports of conjunctival lesions being misdiagnosed as chalazion and chronic follicular conjunctivitis.[10,11] Longstanding rhinosporidiosis of the ocular surface can result in scleral thinning and staphyloma formation and pose a challenge in management.[12,13] Nine cases (26.4%) in the present series had associated scleral thinning and/or staphyloma formation. They needed scleral patch graft for further management. The detailed management has not been discussed because it is out of the scope of the present article.

The next most common site of involvement was lacrimal sac, and it was found in 32.3% of cases of the present cohort. Lacrimal sac lesions commonly present with a mass in the medial canthal region along with watering and are readily misdiagnosed as PANDO with mucocele.[8,14] The rhinosporidial granuloma can be seen filling up the inferior turbinate during a routine preoperative nasal endoscopic examination. In case the rhinosporidiosis is limited only to the lacrimal sac, the sporangia can be seen regurgitating from the puncta on pressure over the sac or while performing a lacrimal sac irrigation. The sac wall might appear eroded while performing a DCR and provide some clue toward the underlying infection. The final diagnosis can be made by opening up the sac wherein the sac would be found filled up with fleshy polypoidal granuloma.

In the present series, too, more than half (54.5%) of the cases of lacrimal sac rhinosporidiosis were misdiagnosed as PANDO, while 3 (27.2%) had already undergone a lacrimal sac surgery elsewhere. One case was misdiagnosed as lacrimal sac neoplasm. Similar rates of misdiagnosis have been reported by other authors, too.[8,14,15] Many of these cases have associated nasal involvement which can be diagnosed on preoperative nasal endoscopy, and the diagnosis can be reconsidered. This highlights the importance of a routine nasal endoscopy in all cases of nasolacrimal duct obstruction before planning surgery. At least three (27.2%) of our cases with lacrimal sac involvement had an abnormal DNE, while one of these had a nasal mass in the inferior turbinate. While most of the authors describe DCT as the preferred technique for lacrimal sac rhinosporidiosis, a technique of modified DCR has been proposed by Nurruddin *et al*. to prevent postoperative bothersome epiphora.[8] Here, the posterior sac flap is completely excised, while the anterior flap is trimmed to leave a small stump. The area around the common internal ostium is packed with 5% povidone iodine-soaked gauze for 2 min to prevent any recurrence. The only other paper on this modified technique is a case series of 13 patients published by Bothra

et al.[14] They, however, used the 5% povidone-iodine for 5 min and additionally placed a nasal pack soaked in povidone-iodine going up to the ostium for 12 h postoperatively. The present study is only the third such article where a modified DCR was performed for lacrimal sac rhinosporidiosis. Because rhinosporidiosis primarily affects the mucosal surface, eyelid dermal involvement is quite rare, and very few such cases have been reported. In a series by Mithal *et al*., there were 11 cases of eyelid rhinosporidiosis.[7] Also, we had just three cases of eyelid involvement in our series. Because the characteristic reddish lesion with typical yellowish dots on the surface is not seen in cutaneous rhinosporidiosis, they are most likely to get misdiagnosed as cutaneous neoplasm. Two of our cases of eyelid rhinosporidiosis were diagnosed as benign eyelid tumor (neurofibroma and lymphangioma) too. While one case was managed by debulking, two cases required excision of the mass with eyelid reconstruction. There are few other rare cases of the eyelid and cutaneous rhinosporidiosis misdiagnosed as neoplasm, too.[16,17]

We had one patient with orbital rhinosporidiosis with extensive involvement in adjacent nasal cavity, sinuses, and nasopharynx. An extensive PubMed search revealed only one such case reported in the literature.[18] Our patient was a 20-year-old male with a chronic course, whereas the case reported by Chakraborty *et al*. presented with features similar to orbital cellulitis.[18] Multispecialty approach was adopted for this patient where open excision of the orbital mass by an orbital surgeon was coupled with endoscopic clearance of sinonasal masses in the same sitting. This was followed by long-term postoperative dapsone therapy for 6 months.

Rhinosporidiosis most commonly affects the mucosal surfaces where it gets lodged after minor trauma. Nose, nasopharynx, paranasal sinuses, conjunctiva, and genitalia are the commonly reported sites. The patients very commonly give a history of bathing in ponds where these parasites find a conducive environment to thrive and grow.[19] They are thought to get carried there by grazing cattle. The infection can also be acquired by inhaling contaminated dust.[20] A rare case of disseminated rhinosporidiosis involving multiple body sites has also been reported.[21] Although the disease is endemic in the Indian subcontinent, Sri Lanka, Bangladesh, and Nepal, there are reports of cases from the Western world as well.[19]

Rhinosporidial lesions are notorious for recurrences. Povidone iodine which causes metabolic inactivation of the endospores and cauterization which causes their thermal killing is effective in preventing recurrences.[22] Oral dapsone (diaminodiphenyl sulfone) causes maturation arrest of the sporangia and accelerates their degeneration. It has been reported to be effective in preventing recurrences and subcutaneous spread.[23] Although there is no fixed protocol, authors have used it in a dosage of 100 mg once or twice daily for 3 months to 1 year.[24] The drug should be used after ruling out allergy and G6PD deficiency. We used oral dapsone in two of our cases for 3 and 6 months, respectively. One of these had extensive orbital and sinonasal involvement, while other had a recurrence after DCT. Both the patients did not have any recurrence at their last follow-up. Recently, a case of disseminated rhinosporidiosis in an immunocompromised individual resistant to dapsone treated with multidrug therapy consisting of cycloserine, dapsone, and ketoconazole has been reported with good response.[25] The use of dapsone should be limited to recurrent cases and those having extensive sinonasal involvement.

The present study is a compilation of the whole spectrum of orbital and adnexal rhinosporidiosis, which has not been done before. However, a limited sample size, retrospective design, and involvement of multiple oculoplastic surgeons in the management are some of the limitations of the present study.

Conjunctiva is the most common site for adnexal rhinosporidiosis, and this is followed by lacrimal sac. The characteristic conjunctival lesions are reddish mulberry-like with pinhead-sized yellowish dots over the surface. Longstanding conjunctival lesions can have associated scleral thinning or an underlying staphyloma. While conjunctival lesions respond well to complete excision with cauterization of the base, lacrimal sac lesions can be managed by a modified DCR. Eyelid rhinosporidiosis can mimic an eyelid mass and should be considered in differential diagnoses in suspicious lesions, especially in endemic areas. Oral dapsone can be considered an adjunct for extensive and recurrent lesions.

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Conflicts of interest

There are no conflicts of interest.

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