

Case Report

Ocular Rhinosporidiosis - A Case Report

Dr. Reeta Dhar¹, Dr. Jyotsna Sahai², Dr. Priyanka Gaikwad², Dr. Shilpi Sahu¹

¹Professor, ²Resident,
MGM Medical College, Kamothe, Navi Mumbai.

Corresponding Author: Dr. Jyotsna Sahai

ABSTRACT

Rhinosporidiosis is a chronic localized granulomatous disease caused by *Rhinosporidium seeberi*. Infestation of rhinosporidiosis in the eye and adnexa is termed oculosporidiosis. In such cases, conjunctival mucosa is mostly involved. A case of ocular rhinosporidiosis diagnosed by the authors is described here.

Keywords: Rhinosporidiosis, Ocular, Conjunctiva.

INTRODUCTION

Rhinosporidiosis is a chronic granulomatous infective disorder caused by *Rhinosporidium seeberi*, an aquatic protistan parasite belonging to a clade, Mesomycetozoa. Its first case was described as a nasal polyp by Guillermo Seeber from Buenos Aires in 1900. ⁽¹⁾ *Rhinosporidium seeberi* was previously considered as a fungus, but now it has been classified under Mesomycetozoa.

Clinically rhinosporidiosis presents as a polypoid soft tissue mass, often pedunculated. Nose and nasopharynx are most commonly involved in it (more than 70% of cases). Ocular lesions, particularly of conjunctiva and lacrimal sac account for about 15% of cases. Rare sites of this disorder include lips, palate, uvula, maxillary antrum, epiglottis, larynx, trachea, bronchus, ear, scalp, vulva, penis, rectum and skin. Rarely disseminated infections are also reported, involving the limbs, trunk and viscera. Brain involvement may lead to fatality. ⁽²⁾

Rhinosporidiosis is endemic in India, Sri Lanka, South America, and Africa. Most of such cases occur in persons

from or residing in the Indian subcontinent. In addition to man, the disease is known to occur in cats, cattle, dogs, ducks, goats, horses, mules, parrots, and swan. ⁽³⁾

It is difficult to culture and isolate the fungus by microbiological methods, though it can be diagnosed by routine hematoxylin and eosin stain, also by special stains such as Gomori's Methenamine Silver (GMS), Periodic Acid Schiff (PAS) and Mucicarmine.

CASE REPORT

A young girl aged 12 years presented with swelling in the left upper palpebral conjunctiva. A month back patient had trauma to the left eye. She did not have any complaints of tearing, drainage, blurring of vision or diminution of vision. There was no history of close contact with animals and no significant past history related to the eye. On examination, a pedunculated lesion arising from the left upper palpebral conjunctiva was observed, not involving the cornea or underlying structures. A clinical diagnosis of papilloma / granuloma was made and surgical excision of the swelling

was performed for histopathological examination.

HISTOPATHOLOGICAL EXAMINATION:

Gross examination - specimen received was a single bit of tissue, soft to firm, grey brown in colour measuring 0.5 cm in length. Microscopic examination - study of hematoxylin and eosin stained sections revealed tissue lined by stratified squamous epithelium. The underlying areas showed fibrocollagenous tissue along with lymphoplasmacytic inflammatory cell infiltrate, occasional neutrophils and numerous congested blood vessels. Also seen were few areas showing large thick

walled sporangia containing numerous endospores of Rhinosporidia.

Upon visualizing endospores of rhinosporidia, special stains were used to confirm the diagnosis.

Periodic Acid Schiff (PAS) highlighted the thick walls of the sporangia in various stages of the life cycle. Mature sporangia revealed a surface pore with release of spores. Gomori Methenamine Silver (GMS) stained the wall of the mature sporangium.

Mucicarmine stain showed positivity by staining of the sporangia and spores. It stained the sporangia, which helps in differentiation with *Coccidioides immitis* as *Coccidioides* doesn't stain with mucicarmine stain. ⁽⁴⁾

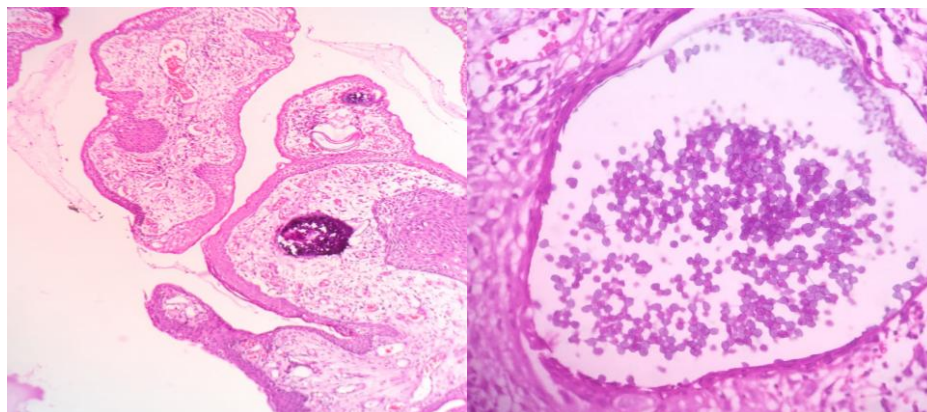


Fig 1 – H and E stain – (a) showing stratified squamous epithelial lining along with sporangia and thick walled cysts, (b) higher magnification showing sporangia and spores.

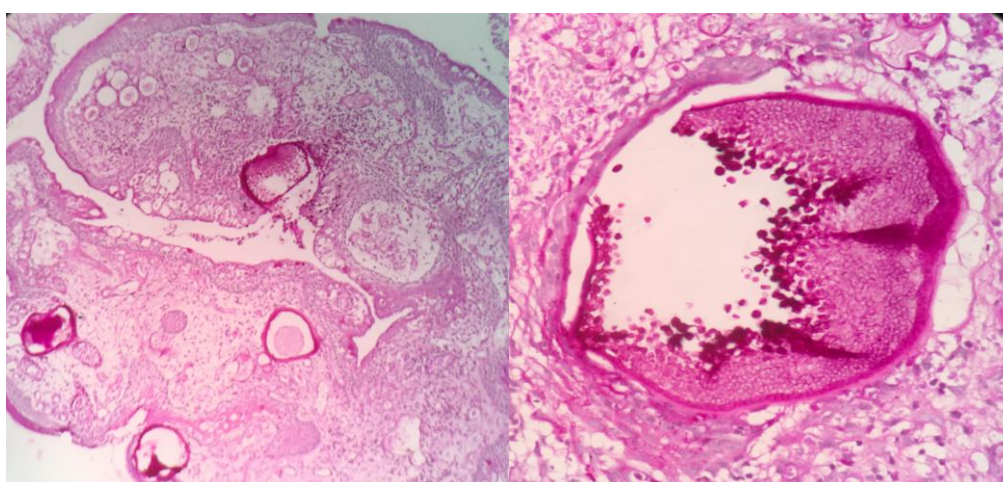


Fig 2 – Periodic Acid Schiff stain – (a) showing multiple cysts and sporangia in varying stages of maturation, (b) higher magnification showing a thick walled cyst with sporangia.

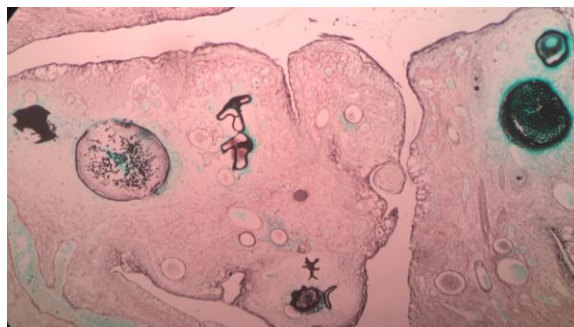


Fig 3 – Gomori's Methenamine Silver Stain (GMS) (10X view) – showing lining epithelium of the lesion along with multiple sporangia and endospores in varying stages of maturation.

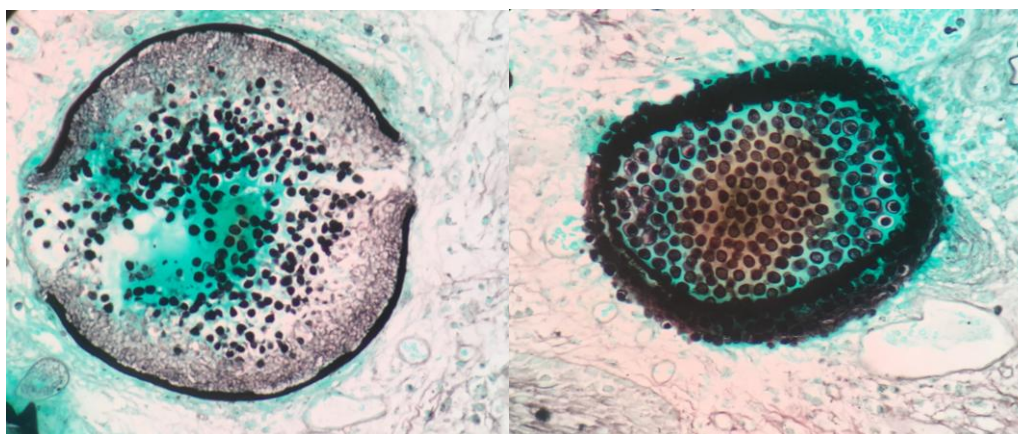


Fig 4 - GMS Stain (40X view) – showing sporangia with endospores in varying stages of maturation.

Based on the above findings and the special stains used, the final diagnosis of Oculo Rhinosporidiosis was given.

DISCUSSION

Rhinosporidiosis is an infective chronic disease which was first observed in Argentina, Latin America over a century ago. Though its prevalence is noted all over the world, in more than 90 countries, it is largely endemic in Indian sub-continent. (5) Though rhinosporidiosis has been reported from over 90 countries, it is usually found in the tropics, being endemic in south India, Sri Lanka, parts of East Africa, and South America. Cases of oculosporidiosis have also been reported from Nepal. (6) Human infection is presumed to occur due to contact of traumatized epithelium with contaminated water and the highest incidence of cases is reported among river - sand workers. (7)

Oculosporidiosis is found among 10-15% of cases of rhinosporidiosis. Conjunctiva is the most commonly affected

tissue (77.6%), lacrimal sac (26%) with or without conjunctival involvement. (8) In this case also it was found that the conjunctiva was involved, to be termed as oculosporidiosis.

Ocular rhinosporidiosis in India affects conjunctiva (69%), lacrimal sac (24%), canaliculi (4%), lids and sclera (4%). (9) The mode of infection and transmission is thought to be water borne, the natural host of this aquatic parasite being fish and amphibians. According to Arseculeratne et al., the commonest source of infection (84%) is lake water followed by rivers (11%) and domestic well water (5%). (10)

Typical polypoid appearance of rhinosporidial lesions helps in correct preoperative diagnosis. But atypical presentations may cause confusion with soft tissue tumours or papillomas. (7) As can be seen in our case, a clinical diagnosis of papilloma or granuloma was made which was thought to have developed post trauma.

The definitive diagnosis of rhinosporidiosis is by histopathological

examination and the organism is always found in the lesion, as was seen in our case also. Rhinosporidial lesions have a distinctive morphology showing polypoid fibroconnective stroma containing globular cysts. Each of these cysts represents a thick-walled sporangium containing numerous "daughter spores" in different stages of development. The stroma contains a vascular fibroconnective tissue and inflammatory infiltrate. ⁽¹¹⁾ Commonly invasive mycoses produce Splendore-Hoeppli reaction characterized by well-developed eosinophilic infiltration in the infected tissue. But rhinosporidiosis is characterized by absence of this reaction, and tissue infiltrate is almost devoid of eosinophils, which were absent even in our case where only a lymphoplasmacytic infiltrate was noted. ⁽¹²⁾ *R. seeberi* is stained by periodic acid-Schiff (PAS), Gomori's methenamine silver, and mucicarmine. ⁽¹¹⁾

Thick-walled sporangia and endospores stain positively with various special stains like PAS, mucicarmine (as used in our study), Gomori's methenamine-silver, Grocott's stain, etc. ⁽⁴⁾⁽⁷⁾⁽¹³⁾ Mucicarmine stain is particularly helpful in differentiating *Coccidioides immitis* as sporangia and spores of this organism do not stain positively. ⁽⁴⁾ Coccidiomycotic lesions can cause confusion with rhinosporidiosis during cytological as well as histopathological evaluation, as the former has similar mature stages represented by large, thick-walled spherical structures containing endospores. But distinction can also be made by H and E stain as intra-sporangial endospores of *R. seeberi* which are larger and more numerous in comparison to those of *C. immitis*. ⁽²⁾

Identification of endospores is often difficult from epithelial cells of the respiratory site, particularly from nasopharynx. Residual mucoid sporangial material around the endospores, referred to as a "comet" form by Beattie, can cause confusion with large nuclei and residual cytoplasm of the epithelial cells. PAS stain is particularly helpful in this setup as

endospores are PAS positive in comparison to the negative staining of epithelial cells. ⁽²⁾

CONCLUSION

Rhinosporidiosis is a very common infection in India with conjunctival ocular rhinosporidiosis being commonest form of ocular rhinosporidiosis. Its clinical presentations may vary and it is best diagnosed by histopathological examination and use of special stains.

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