

Case Report

Oculosporidiosis - Masquerading as a Squamous Papilloma - A Report of Two Cases from Non-Endemic Region

Maryem Ansari^{1*}, Karuna Gupta^{2*}, Ajay Yadav^{2*}, Pankaj Sharma^{2**}, Abid Shamshad³¹Assistant Professor, ²Professor,
*Department of Pathology, **Department of Ophthalmology,
SMS Medical College, Jaipur.³Consultant, Sahai Hospital, Motidungri, Jaipur.

Corresponding Author: Maryem Ansari

*Received: 10/02/2016**Revised: 03/03/2016**Accepted: 09/03/2016*

ABSTRACT

Rhinosporidiosis is an infectious disease affecting the mucosal surfaces. It is not rare to find the disease in areas where it is endemic. But the sporadic cases are rare. We report two cases of rhinosporidiosis from Rajasthan region, which is a non-endemic region for this infection. Another unique feature of both these cases is the site of presentation. Both the cases presented with involvement of the eye which is not the commonest site of infection for the organism.

Keywords: rhinosporidiosis, ocular, squamous papilloma.

INTRODUCTION

Rhinosporidiosis is a chronic granulomatous infection of the mucous membranes of specific sites (nasal, oral, ocular and rectal) caused by the organism rhinosporidiosis seeberi. This organism was earlier thought to be a fungus but now it has been classified in a new class "mesomycetozoa" which means intermediate of "myceto" (fungus) and "zoa" (animals). ⁽¹⁾ Most commonly the patients present with nasal pathology. Only a fraction of cases present with complaints at other sites. Oculosporidiosis (rhinosporidiosis of the eye) is one of the unusual sites for presentation. We report two cases of rhinosporidiosis of eye diagnosed at our institute.

CASE REPORT

Case no. 1

A healthy young male came to the Ophthalmology department with the

presenting complaints of small mass on the right eyelid. He had no other complaints related to vision. There was no associated watering or redness. The patient was diagnosed as a case of lid papilloma and was operated for the same.

Case no. 2

A 10 years old child came to the outpatient department with mass in left eye. There was no other complaint except for slight limitation in the field of vision. Examination revealed red fleshy mass in the medial part of the bulbar conjunctiva in the left eye. A diagnosis of conjunctival papilloma was made.

Both patients were local natives. There was no history of visit to any coastal or endemic region in both the cases. Lacrimal ducts and opening were normal in both the cases. There was no discharge, lymphadenopathy or photophobia in either of the case.

The mass was surgically excised in both the cases and sent for histopathology examination. The tissue was processed entirely. Sections were stained using the hematoxylin and eosin stains. Microscopic examination revealed characteristic features of rhinosporidiosis comprising of multiple sporangia in various stages of development. The mature sporangia were filled with spores also known as nucleated basophilic endoconidia/ endospores (Fig. 1). At many places the sporangia were seen bursting with release of the spores.

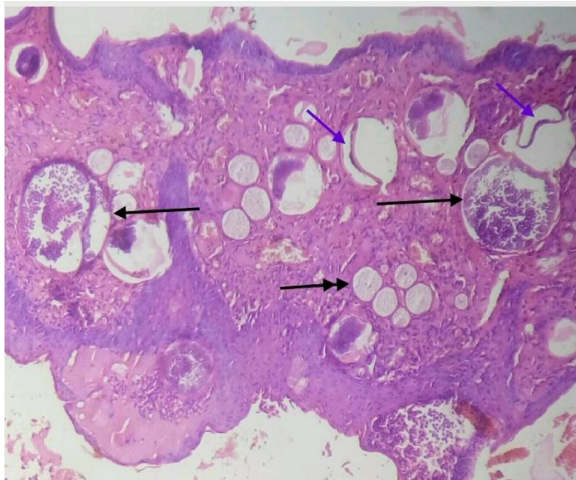


Figure 1: Hematoxylin and Eosin stain showing sporangia in various stages of development. Lining squamous epithelium shows hyperplasia. Single black arrow - mature sporangia filled with spores. Double black arrow - immature sporangia. Single blue arrow - collapsing sporangia.

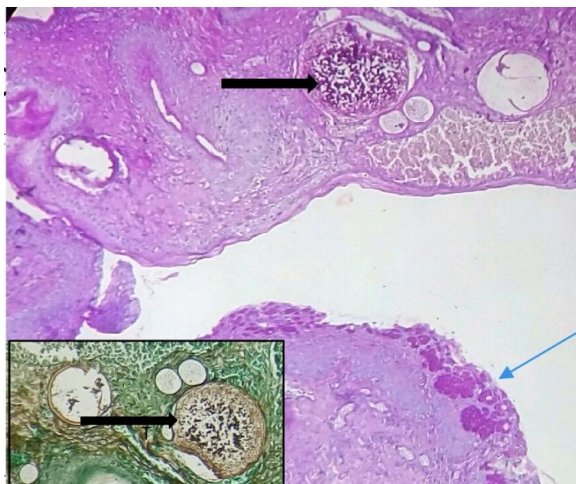


Figure 2: PAS positive spores are seen along with positively stained goblet cells of the conjunctival epithelium (blue arrow). Inset - GMS stain showing positively stained spores.

In the first case an area showed ulcerated squamous epithelium covered with plaque with underlying tissue showing mixed inflammatory cells. In

second case squamous epithelium revealed hyperplasia, papillomatosis and invagination forming pseudocyst in addition to above mentioned findings. Apart from the usual hematoxylin and eosin stains, special stains like Periodic acid Schiff (PAS) and Gomoris methenamine silver (GMS) stain were also performed. The spores stained positive for PAS stain and GMS (Fig.2). The thick chitinous wall of the sporangia also stained positively for both the stains.

DISCUSSION

Rhinosporidiosis is a chronic granulomatous inflammatory disorder, worldwide in distribution but more commonly seen in South Asian countries like India, Srilanka and Nepal. (2,3) Sporadic cases have been reported from other countries also. (4-6)

Usually, either the cases are reported from endemic regions or the patients have history of visit to any such region. Sporadic cases are rare, with only few reports in literature. Our cases are unique because both the patients are natives of Rajasthan, where the climatic conditions are dry and arid as opposed to the humid atmosphere favourable for the growth of rhinosporidiosis. Moreover neither of them had any history of visit to any such region.

Males are more commonly affected. The sex predilection can be accounted by the occupational exposure. Though some authors do not report any sex predilection, reporting insignificant differences in the incidence in males and females, occasional authors have even reported a female preponderance. Children, adolescents and young adults are more commonly affected because of their increased outdoor activity.

The mode of spread of the organism is through stagnant water bodies. By taking bath or through working, transmission of the organism occurs through the mucosa. The organism most commonly affects the mucosa of the nasal

cavity, nasopharynx, larynx soft palate and buccal cavity. Second most common site affected is the eye and the ocular adnexa with such cases ranging from 9% to 24 % and even higher in certain subdivisions. (2,7)

The structures of the eye involved are as follows in order of frequency of affection - conjunctiva, lacrimal sac, and lids. Our first case presented with lesion at the site which is least commonly encountered site-the eyelids. The presentation was however similar to the usual presentation. It was a red fleshy polypoidal mass. This form of presentation is very common and often misdiagnosed as papilloma. (8) Other unusual forms of presentation of oculosporeidiosis have also been reported such as staphyloma formation and cystic mass formation. (9,10) Ocular rhinosporidiosis may also present as recurrent chalazion, chronic follicular conjunctivitis and may even lead to mechanical ectropion. (11-13)

Treatment

The treatment of rhinosporidiosis is surgical excision with cauterisation of the base. Rarely spontaneous regression has been noted. Recurrence is possible because of spillage of endospores in the adjacent mucosa. So the patient is advised to have regular follow-up.

Medical treatment in the form of dapsone has been recommended by some. The mechanism of action is believed to be arresting the maturation of sporangia.

CONCLUSION

This case report brings into light the fact that rhinosporidiosis may be considered as a differential diagnosis in patients presenting with mass in the eye even in non endemic regions. The treatment should be preferably surgical along with medical aid. A close follow up of the patient should be kept as there are high chances of recurrences.

REFERENCES

1. Vilela R, Mendoza L. The taxonomy and phylogenetics of the human and animal pathogen *Rhinosporidium seeberi*: a critical review. *Rev Iberoam Micol.* 2012; 29(4):185-199.
2. Arseculeratne SN. Recent advances in rhinosporidiosis and *Rhinosporidium seeberi*. *Indian J Med Microbiol.* 2002; 20(3):119-131.
3. Shrestha SP, Henning A, Parija SC. Prevalence of rhinosporidiosis of the eye and its adnexa in Nepal. *Am J. Trop Med Hyg.* 1998; 59:231-4.
4. Harissi-Dagher M, Robillard N, Corriveau et al. Histopathologically confirmed ocular rhinosporidiosis in two Canadians. *Can J Ophthalmol* 2006; 41:227-229.
5. Morelli L, Polce M, Pisciolli F et al. Human nasal rhinosporidiosis: an Italian case report. *Diagnostic Pathology.* 2006; 1:25.
6. Kaimbo KW, Parys-Van Ginderdeuren R: Conjunctival rhinosporidiosis: a case report from a Congolese patient. *Bull Soc Belge Ophtalmol* 2008; 309-310:19-22.
7. Moses JS, Balachandran C, Sandhanam S et al. Ocular rhinosporidiosis in Tamil Nadu, India. *Mycopathologia.* 1990; 111: 5-8.
8. Gichuhi S, Onyuma T, Macharia E et al. Ocular rhinosporidiosis mimicking conjunctival squamous papilloma in Kenya - a case report. *BMC Ophthalmology.* 2014; 14:45.
9. Jacob P, Rose JS, Hoshing A et al. Tectonic corneal graft for conjunctival rhinosporidiosis with scleral melt. *Indian J Ophthalmol.* 2011; 59: 251-253.

10. Lavaju P, Arya SK, Kumar B, Upadhaya P: Conjunctival rhinosporidiosis presenting as a cystic mass-an unusual presentation. Nepal J Ophthalmol: NEPJOPH 2010; 2(2):157-159.
11. Mukhopadhyay S, Shome S, Bar PK et al. Ocular rhinosporidiosis presenting as recurrent chalazion. Int Ophthalmol 2012; 1-3.
12. Suh LH, Barron J, Dubovy SR et al. Ocular rhinosporidiosis presenting as chronic follicular conjunctivitis in a contact lens wearer. Arch Ophthalmol 2009; 127(8):1076-1077.
13. Mandal SK, Bhakta A, Mandal A et al. Giant mass conjunctival rhinosporidiosis causing severe mechanical ectropion. J Indian Med Assoc 2012; 110(5): 328-329.

How to cite this article: Ansari M, Gupta K, Yadav A et al. Oculospodidiosis - masquerading as a squamous papilloma - a report of two cases from non-endemic region. Int J Health Sci Res. 2016; 6(4):549-552.

International Journal of Health Sciences & Research (IJHSR)

Publish your work in this journal

The International Journal of Health Sciences & Research is a multidisciplinary indexed open access double-blind peer-reviewed international journal that publishes original research articles from all areas of health sciences and allied branches. This monthly journal is characterised by rapid publication of reviews, original research and case reports across all the fields of health sciences. The details of journal are available on its official website (www.ijhsr.org).

Submit your manuscript by email: editor.ijhsr@gmail.com OR editor.ijhsr@yahoo.com