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A clinicopathological study of rhinosporidiosis in a tertiary care hospital

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ABSTRACT

Background: Rhinosporidiosis is a chronic granulomatous infective disorder that is caused by *Rhinosporidium seeberi*. It usually presents as a soft polypoidal, pedunculated or sessile mass arising from the nasal mucosa. Common sites of occurrence of rhinosporidiosis are nasal cavity and nasopharynx, it can also be found in conjunctiva, larynx and maxillary sinuses.

Methods: A cross-sectional study was conducted in the outpatient department of ENT, at Vinayaka Mission's Medical College and Hospital for a period of 1 year 7 months from October 2011 to April 2013. During the study period, all cases that were diagnosed as rhinosporidiosis by histopathology were included in the study group. The aim of this study is to determine the epidemiology, risk factors, clinical features and evaluation of blood group in patients with rhinosporidiosis in a study group.

Results: Majority of patients in our study were young male adults from low socio-economic strata and from rural area. The common sites involved were the nasal cavity and nasopharynx. It showed an association with blood group O type.

Conclusions: Rhinosporidiosis is an infective disease which is seen in individuals using surface water sources for daily needs. It requires careful clinical evaluation and diagnosis. Patients in high risk group with suspicion should undergo surgical excision with electrocautery. Careful follow up is essential for early diagnosis of recurrence.

Keywords: Rhinosporidiosis, *Rhinosporidium seeberi*, Nasal mass, Surgical excision, Dapsone therapy

INTRODUCTION

Rhinosporidiosis is a chronic granulomatous infective disorder commonly affecting the mucous membranes of nose, nasopharynx and less commonly other sites. The etiological agent of rhinosporidiosis is *Rhinosporidium seeberi*. The causative pathogen is widely deemed to be a Fungus though its precise taxonomy is still a matter of debate.

The first case was described by Guillermo Seeber from Buenos Aires in 1900. The causative organism was initially considered as a fungus, and Ashworth in 1923 described it's life cycle establishing the nomenclature *Rhinosporidium seeberi*.²

The disease is endemic in India and Sri Lanka, however sporadic cases have also been reported from United States of America, South America, Italy, Iran and Turkey.³⁻⁵

The present state of research concerning the taxonomy of *Rhinosporidium seeberi*: whether it is a prokaryotic *Cyanobacteria* or a eukaryotic parasite is controversial. Recent studies found a small subunit sequence of r DNA to be similar to that of members of the *Dermocystidium*

genus: aquatic protistan fish parasites that belong to the DRIP clade.⁶

The disease has been isolated not only from humans but also from other animals such as cattle, buffaloes, horses, mules. Mode of spread also remains debatable, the more acceptable being that it spreads from sand and water especially stagnant water sources like wells, pond, tanks etc in endemic areas. Nasal rhinosporidiosis presents as pedunculated, 'strawberry' like, bulky, friable soft tissue mass and is confirmed by histopathological examination.

Conjunctiva and Lacrimal sac can also be involved. Rare sites of involvement are lips, palate, uvula, maxillary antrum, epiglottis, larynx, trachea, bronchus, ear, scalp, vulva, penis, rectum and skin. Rarely, disseminated infections are also reported, involving limbs, trunks and viscera. Treatment consists of surgical excision but it presents with high recurrence rates.

There can be occasional dissemination from the initial focus which is most commonly seen in upper respiratory sites. Rarely, spontaneous regression of rhinosporidial nasal masses has been documented.^{8,9}

The present study was conducted to establish clinical and epidemiological profile of the patients who present with rhinosporidiosis.

METHODS

A cross-sectional study was conducted in the department of ENT of Vinayaka Mission's Medical College and Hospital, Karaikal, India, for a period of 1 years 7 months from October 2011 to April 2013. A total of 50 patients were included in our study. During the study period, all cases that were diagnosed as rhinosporidiosis by histopathology were included in the study group. A detailed history was taken and clinical examination was carried out in all patients with suspected Rhinosporidial lesions. Routine blood examination was carried out including ABO blood grouping and Rh typing in all the cases. The excised soft tissue masses were sent for Histopathological examination using haematoxylin and eosin staining. In some cases special stains such as Gomori methamine, periodic acid Schiff (PAS) and mucicarmine were used. Evidence of thick-walled sporangia containing numerous endospores in a background of a fibrovascular stroma on microscopic examination of the excised specimen established the diagnosis of rhinosporidiosis.

RESULTS

During the 19 months of study, rhinosporidiosis constituted (16%) of all nasal masses that were operated in our institute. Incidence of rhinosporidiosis was found to be about 30 cases per year. Among the 50 cases, 42 patients had rhinosporidiosis lesion restricted to the nasal cavity and 8 patients had nasopharyngeal extension. 15

(30%) patients had recurrence of rhinosporidiosis and had initially been operated outside our institution. Patients presented with symptoms of nasal mass, nasal obstruction, nasal discharge and epistaxis. All the cases were confirmed by histopathological examination.

Table 1: Sex and age distribution of rhinosporidiosis.

Age group	Male	Female	Total
0-10	3	0	3
11-20	12	6	18
21-30	13	4	17
31-40	5	3	8
41-50	1	1	2
51-60	1	0	1
>60	1	0	1
Total	36	14	50

In this study it was found that males were predominantly affected than females, male:female ratio being 2.5:1. Most of the patients in our study belonged to the 11-20 and 21-30 years age group.

Table 2: Occupation of the patient.

Occupation	Number
Farmer	20
Student	15
Fishermen/fisherwomen	7
Housewife	3
Factory worker	5

Table 3: Residential status of the patient.

Residential status	Number
Rural	41
Semi-urban	7
Urban	2

Table 4: Bathing history of the patient:

Bathing history	Number
Irrigation canals	3
Rivers	5
Ponds	36
Wells	6

Table 5: Socioeconomic status.

Socioeconomic status	Number
Low	38
Middle	12
High	0

Majority of the patients were farmers (40%) followed by students, living in rural areas (82%) and were from low socio-economic status (76%). Majority of the patients had history of bathing in ponds and other surface water sources.

Table 6: Presenting features.

Presenting features	Number
Mass in the nose	36
Nasal obstruction	33
Bleeding from the nose	14
Nasal discharge	12
Change in voice	2
Headache	3

Majority of patients presented with the feature of nasal mass (36; 72%). Out of 50 patients, 33 patients (66%) presented with nasal obstruction, 14 patients (28%) presented with nasal bleeding and 12 patients (24%) presented with nasal discharge. Change in voice and headache were less common. Predominantly patients presented with complaints of nasal mass and nasal obstruction, majority of them (32 patients) presenting with more than one complaints.

Table 7: Site distribution.

Site distribution	Number
Lateral wall	22
Nasal septum	18
Nasal floor	3
Nasopharynx	1
Multiple sites	6

Lateral wall of nasal cavity was the most frequent site of attachment of rhinosporidiosis, found in 22 patients (44%) in our study followed by the nasal septum.

Table 8: Number of lesions.

Number of lesions	Number
Solitary	43
Multiple	7

Majority of the lesions were solitary in nature in our study. Majority of the patients had unilateral mass. Majority of the patients belonged to the blood group O.

Table 9: Laterality.

Laterality	Number
Unilateral	46
Bilateral	4

Table 10: Incidence of Rhinosporidiosis in relation to blood group of the patient.

Blood group of the patients	Number
A	11
В	1
AB	2
0	36

Surgical excision of the nasal masses was carried out, nasal endoscope was used to locate all the lesions and also to aid in cauterizing the base. Electrocautery was used to cauterize the base of the lesion. Dapsone was given post-operatively for a duration of 6 months, orally in the dose of Tablet Dapsone 2 mg/kg/day. Patients were advised to stop using surface water sources for their daily needs and instead use tap water for bathing. Most of the cases were followed-up for a minimum period of one year. Diagnostic nasal endoscopy was done in all the patients during follow-up visits. 4 patients showed evidence of recurrence on follow-up and were treated again by surgical means.

DISCUSSION

Rhinosporidium seeberi was first described as sporozoan by Malbran in 1892 in Argentina. Seeber classified it as protozoan in 1900. Ashworth thought it to be phycomycetes and proposed the name Rhinosporidium seeberi. Some authors have also put it under a new class of aquatic protistans called mesomycetozoea along with other aquatic parasites that cause similar infections in amphibians and fish. However, recent studies suggest a waterborne cyanobacterium Microcystis aeruginosa as the causative organism of rhinosporidiosis.

The taxonomy of the causative organism is unclear. It shows morphological features resembling those of fungi and protozoa. Although most microbiologists had initially considered it a fungus as fungal stains like methamine silver and PAS could stain the wall of the organism but culture of R. seeberi had been unsuccessful in all artificial media; however it could be maintained through it's life cycle in tissue cultures. Ahluwalia hypothesized that the causative organism was not a fungus but a prokaryotic cyanobacterium Microcystis aeruginosa, based on the findings that this organism was isolated from both the clinical specimen of patients and pond water samples where they bathed. 3,12,13 Herr et al however, through analysis of 18s small subunit ribosomal DNA groups, concluded that R. seeberi was related to a group of fish parasites referred to as the DRIP clade (Dermocystidium rosette agent, icthyophonus and psorospermium). The biological agent has a mature stage that consists of large thick walled spherical structures called sporangia containing smaller "daughter cells" called sporangiospores and it can be visualized with fungal stains like Gomori methamine silver and PAS, as well as with standard haematoxylin and eosin staining.⁶

The presumed mode of infection from natural aquatic habitat of *R. seeberi* is through traumatized nasal epithelium. The occurrence of rhinosporidiosis in river sand workers in India and Sri Lanka is partially relevant to such a mode of infection, through abrasions caused by sand particles with the pathogen in putative habitat-ground water. "Auto-inoculation" was considered as the explanation for the occurrence of satellite lesions adjacent to granulomas especially in the upper respiratory sites

and for local spread.² Spillage of endospores from the lesion after trauma or surgery is thought to be followed by autoinoculation through the adjacent epithelium. Majority of the cases are sporadic. An isolated outbreak of ocular and nasal rhinosporidiosis was reported in Serbia.⁸

Rhinosporidiosis is common among sand workers, paddy cultivators and people bathing in stagnant muddy water. As the medical college in which the study was conducted is situated in a rural area, most of the patients coming to hospital are poor villagers, fishermen and agriculturists who are accustomed to take bath in surface water sources and are prone to mucosal injuries by sand or dust. Direct man to man transmission is not known. Being a coastal area, the region has many surface water sources developed as a result of small riverlets joining the sea and also because of back waters which were used as water source by rural population. Fishermen and other villagers in this region have a habit of getting into surface water sources for catching fish using various techniques as fish forms an important part of diet of people living in this region. Students were also found to be affected as they have a habit of swimming in surface water sources.

In this study it was found that males were predominantly affected than females. This could be due to the habit of males taking bath in the ponds after washing their cattle. Fewer females used to take bath at ponds and other surface water sources and most of them used to take bath at home using tap water leading to lower incidence amongst females. This observation is similar to the one noticed in studies conducted by Lakshman et al and Mahmud et al. ^{15,16}

Most of the patients in our study were in the 2nd and 3rd decades of life, the reason could be that the Individuals in this age group used to take the cattle to ponds and also swim in same water source. The disease is seen in all age group but is more often in early decades of life. Similar results with respect to age incidence of rhinosporidiosis were found in studies by Satyanarayana C and Mahmud et al. ^{16,17}

A vast majority of patients (48%) presented with symptoms of both nasal mass and nasal obstruction. Most of the patients had more than one symptoms. Presence of nasal obstruction together with nasal mass were the most common co-existing symptoms, rather than epistaxis which was similar to the findings of Iqbal and Dani. ¹⁸

Lateral wall of nasal cavity was the most frequent (29 patients, 58%) site of attachment of rhinosporidiosis in our study followed by the nasal septum. In 6 cases rhinosporidiosis was found to be attached to multiple sites. In one of the patients rhinosporidiosis was attached to the nasopharyngeal mucosa.

Majority of the patients in our study had a unilateral nasal mass and the lesions were solitary in nature. Nasal endoscopy was done to detect the exact number and site of attachment of rhinosporidiosis lesions in the nasal cavity and nasopharynx.

Rhinosporidiosis of nasal passage usually appears as a bulky, friable, granular, polypoid mass, red in color with multiple yellowish pin head-sized spots representing underlying mature sporangia. This gross appearance, though distinctive, is not diagnostic. Definitive diagnosis of rhinosporidiosis depends upon identification of the pathogen in its diverse stages in excised tissue by histopathological examination. ²¹

Routine hematological evaluation in our study did not reveal any significant abnormality. Majority of the cases had a normal total leucocyte count. There was no evidence of eosinophilia. Similar experience has also been reported in other studies. 9,20,22,23 ABO blood grouping of the patients revealed that 72% of the patients belonged to blood group O, followed by 22% belonging to blood group A. Few studies have shown that the highest incidence of rhinosporidiosis was in group O. 24 But, according to some authors blood group distribution is too variable to draw any conclusion. 25

The mainstay of treatment is surgical excision of the lesion. Wide surgical excision that is advised cannot be done in most of the cases because it can result in complications such as septal perforation, excessive crusting, atrophic rhinitis, haemorrhage. Total excision of the lesion, followed by electrocautery of the base, is the recommended mode of treatment. Recurrence may occur due to spillage of endospores in the surrounding mucosa during removal.²¹ Good results without recurrence have been reported, following the use of endoscope during surgery.¹⁴ Recurrences are common and the only drug useful in reducing recurrence is dapsone. Dapsone (4, 4 diamino diphenyl sulphone) arrests the maturation of sporangia and promotes fibrosis in the stroma, when used as adjunct to surgery.²⁶

Rhinosporidiosis is an infective condition which is commonly seen in 2nd and 3rd decade of life. It is seen in people belonging to low socio-economic group and those living in rural areas being more common in males, in individuals who take bath in surface water bodies. In most of the cases, lesions of rhinosporidiosis are restricted to nasal cavity hence present with nasal complaints such as nasal mass, nasal bleeding and nasal obstruction. Nasal endoscopy has to be done in all the cases to detect the site of attachment and the number of lesions. Laryngoscopy is advised in order to rule out rhinosporidiosis lesions in other sites like larynx, laryngopharynx. Surgical excision with electrocautery of the base is preferred to reduce recurrence. Post-operative medical therapy with Dapsone is known to have a limited role in reducing the chances of recurrence. Repeated follow-up of the patients for early detection of lesions is essential. Certain precautions, like avoidance of use of surface water for bathing and other domestic purposes also helps in reducing the chances of recurrence. Washing of cattle and utilization of water from same water sources for human needs, should be discouraged. In general, improvement in sanitation and general hygiene, making people in rural areas accessible to piped water supply along with imparting proper health education to the high risk groups residing in rural areas of coastal India can reduce the incidence of rhinosporidiosis in the high risk rural population.

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