

# A Retrospective Epidemiological Study of Rhinosporidiosis in a Rural Tertiary Care Centre in Pondicherry

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## ABSTRACT

**Introduction:** Rhinosporidiosis is an age old endemic scourge which has affected various parts of the world, most notably India and Sri Lanka. Although a large body of literature exists regarding this problem, postoperative recurrence rates continue to vary, and the effect of public health education in the eradication of this disease has not been taken into account.

**Aim:** The aim of this study was to find out the site, presentation and postoperative recurrence rate in patients with rhinosporidiosis.

**Materials and Methods:** This was a retrospective study involving operated cases of rhinosporidiosis in the Department of Ear, Nose and Throat of a rural tertiary care referral centre over a period of 3 years. The behavioural pattern of the disease, its epidemiology, rate of recurrence and outcomes of surgical management were analysed.

**Results:** A statistically significant correlation could be found between male gender and rural residential status and incidence of rhinosporidiosis. Bathing in rivers and ponds were found to be strongly associated with rhinosporidiosis ( $p=0.005$ ). Almost all patients were found to be of low socioeconomic status. Nasal obstruction and epistaxis were the most common presenting complaints.

**Conclusion:** Rhinosporidiosis is strongly associated with male gender, young and middle aged adults, agricultural occupations, rural residential status, a history of bathing in ponds and rivers and a low socioeconomic status. Post-surgical recurrence of rhinosporidial masses can be avoided with careful and complete clearance of the mass and cauterization of the base. Appropriate and consistent public health education helps to reduce the incidence of rhinosporidiosis in endemic communities.

**Keywords:** Aetiology, Clinical features, *Rhinosporidium seeberi*, Surgical outcomes

## INTRODUCTION

Rhinosporidiosis is an enigmatic disease which has been known to medicine since 1900 following its first description by Guillermo Seeber in Buenos Aires, Argentina, who proposed the infective aetiology for this disease to be a fungus, which was later isolated by Ashworth in 1923, who in turn described the life cycle of the organism and established the nomenclature *Rhinosporidium seeberi* [1]. Rhinosporidiosis is a chronic, granulomatous disease process which is characterized by reddish polypoidal masses which are hyperplastic and friable and may be pedunculated or sessile. *Rhinosporidium seeberi* has been found to have an affinity for the mucous membranes of the nasal cavity and nasopharynx, mainly affecting the anterior nasal septum and vestibule. Extranasal involvement is rare, particularly that involving the lower aerodigestive tract including the tracheobronchial tree. Various other sites have been reported in literature and these include conjunctiva, lips, uvula, palate, trachea, larynx, vagina, penis and bone [2,3].

Hot tropical climates have been found to be the most suitable environment for this organism, and it is hyperendemic in Sri Lanka and Southern India. This disease has been reported from about 70 countries including Europe, North and South America, Brazil, Argentina, Mexico with scattered reports from Columbia, Venezuela, Uganda, Madagascar, Ghana, Iran, Russia and South East Asia [4]. 'Transepithelial infection', which is infection via traumatized epithelium, has been proposed as a probable mode of infection from the natural aquatic environment of this organism. The diagnosis is straightforward when the disease process involves the nasal cavity or the nasopharynx due to the typical gross characteristics of the lesions, but becomes more difficult and is often delayed when extranasal sites are involved. A detailed history of bathing habits and occupational exposure to stagnant water helps to clinch the clinical diagnosis, while histopathological examination of the excised mass confirms it [5-8].

## AIM

The aim of this study was to find out the site, presentation and postoperative recurrence rate in patients affected with rhinosporidiosis in the Pondicherry region.

## MATERIALS AND METHODS

This was a retrospective study involving 32 patients of histologically proven rhinosporidiosis treated in the Department of Otorhinolaryngology and Head and Neck Surgery of a tertiary care centre in Pondicherry, South India, over a period of 3 years from January 2012 to December 2014, following approval by the Institutional Ethical Committee. All patients had undergone a detailed history taking and clinical examination which were retrieved from hospital case records and personal interviews. Detailed history included duration of symptoms, personal habits, area of residence, bathing habits and work profile of the patient. Diagnosis was made on the basis of detailed history, clinical examination and a battery of investigations including Plain X-ray Nose and Paranasal Sinuses, CT Paranasal Sinuses (coronal and axial views), Diagnostic nasal endoscopy and histopathological examination. All patients were subjected to routine haematological investigations including blood grouping for operative fitness and underwent endoscopic excision of the mass and electrocautery of the base of lesion under general anaesthesia. Following excision, the entire nasal cavity and nasopharynx were examined for hidden lesions. All excised specimens were sent for histopathological examination and confirmation of the diagnosis. All patients had been followed up for a period of 12 months and were subjected to clinical examination and diagnostic nasal endoscopy at regular intervals at 2 weeks, 3 months, 6 months, 9 months and at the end of 1 year.

## STATISTICAL ANALYSIS

The results were collected and recorded in a computerized proforma and statistically analysed. Statistical analysis was carried out using SPSS version 20.0 (SPSS Inc., Chicago, IL, USA) software with Regression Modules installed. Chi-square test and t-test were applied. The p-values of 0.05 or less were considered statistically significant.

## RESULTS

This study involved 32 cases of histologically proven rhinosporidiosis over a period of 3 years. Of these 32 cases, 18 (56.25%) were male and 14 (43.75%) were females. The male to female ratio was found to be 1.3:1. A statistically significant correlation could be found between male gender and incidence of rhinosporidiosis ( $p=0.03$ ) [Table/Fig-1].

Most cases were found to fall within the 21-50 years age group, with the maximum in the third and fourth decades of life, but no statistically significant relationship could be found between age and incidence of rhinosporidiosis ( $p=0.81$ ). The youngest patient in this series was eight-year-old and the oldest was 74-year-old [Table/Fig-1].

A multivariate analysis of possible aetiological factors was conducted including occupation, residential status, socioeconomic status and bathing history. The patients in this study were mainly farmers, housewives or students, with a few daily wage labourers and factory workers. Although rhinosporidiosis was found most commonly in farmers, housewives and students, no statistically significant correlation could be determined ( $p > 0.05$ ) [Table/Fig-2].

The majority of cases lived in rural areas (59.38%), while the remaining lived in semi-urban (21.88%) and urban (18.75%) areas. A significant relationship was found between rural residential status and incidence of rhinosporidiosis ( $p=0.02$ ).

Almost all patients gave a history of bathing in ponds (59.38%) or rivers (31.25%). Here, again a statistically very significant relationship was seen between bathing in rivers and ponds and the incidence of rhinosporidiosis ( $p=0.005$ ).

Almost all patients were found to be of low socioeconomic status (75%) as compared to 25% of patients who were of middle socioeconomic status, and this was found to be statistically significant ( $p=0.047$ ).

The symptoms with which the patients presented to the out patient department included nasal obstruction, epistaxis, nasal discharge and complaints of nasal mass. Of these, nasal obstruction (40.63%) and epistaxis (28.13%) were the most common presenting complaints among patients with rhinosporidiosis, and these were found to have a strong association ( $p=0.0005$ ) [Table/Fig-3].

On examination, the majority of patients were found to have sessile masses (65.63%) and the primary site of attachment was found to be the inferior turbinate and septum in the nose. 21.88% of patients were found to have primary involvement of the nasopharynx while 18.75% were found to have primary nasal and nasopharyngeal involvement. Neither the gross characteristics of the masses nor the site of primary attachment were found to have any statistical significance ( $p > 0.05$ ) [Table/Fig-3,4].

Aetiology		No. (%)	p-value
Occupation	Farmer	9 (28.13%)	>0.05
	Daily wage labourer	2 (6.25%)	
	Student	8 (25%)	
	Housewife	10 (31.25%)	
	Factory worker	3 (9.38%)	
Residential status	Rural	19 (59.38%)	0.02
	Urban	6 (18.75%)	
	Semi-urban	7 (21.88%)	
Bathing history	Reservoirs	2 (6.25%)	0.005
	Rivers	10 (31.25%)	
	Ponds	19 (59.38%)	
	Tap water	1 (9.38%)	
Socioeconomic status	Low	24 (75%)	0.047
	Middle	8 (25%)	
	High	0 (0%)	

[Table/Fig-2]: Aetiologic characteristics.

Feature		No. (%)	p-value
Symptoms	Nasal mass	5 (15.63%)	0.0005
	Nasal obstruction	13 (40.63%)	
	Epistaxis	9 (28.13%)	
	Nasal discharge	5 (15.63%)	
Gross characteristics	Pedunculated	11 (34.38%)	0.0964
	Sessile	21 (65.63%)	
Site of involvement	Nasal	19 (59.38%)	0.0042
	Nasopharyngeal	7 (21.88%)	
	Nasal and nasopharyngeal	5 (15.63%)	
	Disseminated	1 (3.13%)	
Site of primary attachment	Inferior turbinate	6 (18.75%)	>0.05
	Septum	7 (21.88%)	
	Middle turbinate	1 (3.13%)	
	Middle meatus	1 (3.13%)	
	Inferior turbinate	2 (6.25%)	
	Multiple nasal attachments	2 (6.25%)	
	Nasal and nasopharyngeal	6 (18.75%)	
	Nasopharyngeal	7 (21.88%)	

[Table/Fig-3]: Clinical features.

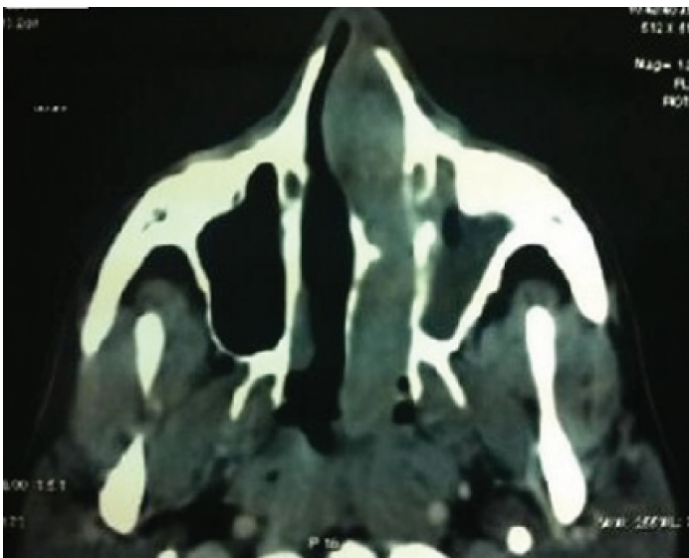
The site of involvement was found to be nasal in 59.38% of patients, nasopharyngeal in 21.88% of patients and both nasal and nasopharyngeal in 15.63% of patients [Table/Fig-5]. One patient in this study presented with disseminated rhinosporidiosis [Table/Fig-6]. Involvement of the nose alone was found to be statistically significant ( $p=0.0042$ ). The ratio of nasal to extra-nasal involvement was found to be 1.5:1. None of the cases showed any enlargement of the regional lymph nodes [Table/Fig-3].

Gender	Age (in Years)								Total
	0 - 10	11 - 20	21 - 30	31 - 40	41 - 50	51 - 60	61 - 70	71 - 80	
Male	3 (9.38%)	2 (6.25%)	3 (9.38%)	5 (15.63%)	4 (12.5%)	0 (0%)	1 (3.13%)	0 (0%)	18 (56.25%)
Female	1 (3.13%)	1 (3.13%)	3 (9.38%)	4 (12.5%)	2 (6.25%)	1 (3.13%)	1 (3.13%)	1 (3.13%)	14 (43.75%)
Total	4 (12.5%)	3 (9.38%)	6 (18.75%)	9 (28.13%)	6 (18.75%)	1 (3.13%)	2 (6.25%)	1 (3.13%)	32 (100%)

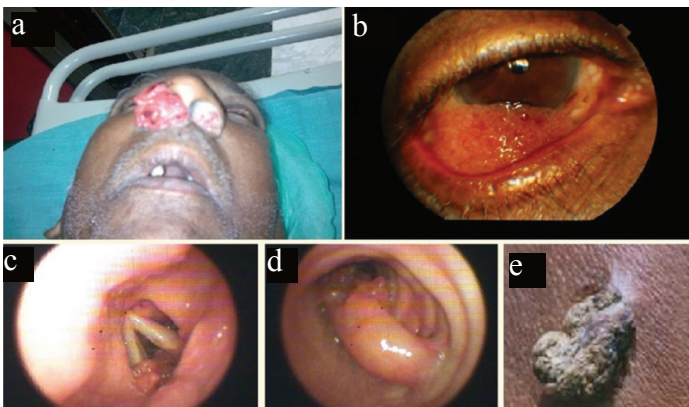
[Table /Fig-1]: Age-gender distribution.  
p-value = 0.817.



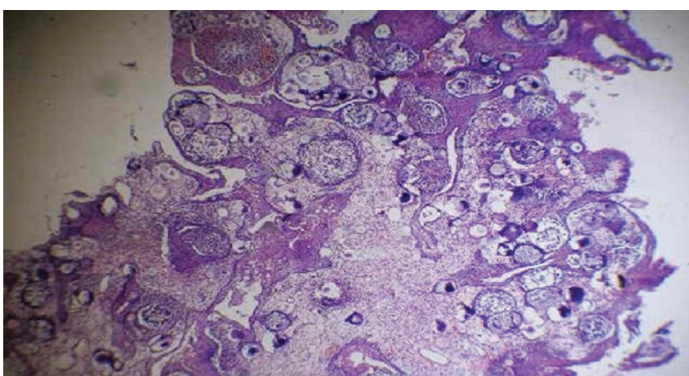
**[Table/Fig-4]:** Showing rhinosporidial mass protruding out through left nasal cavity.



**[Table/Fig-5]:** Showing axial view of plain CT nose and paranasal sinuses with soft tissue density lesion filling left nasal cavity and nasopharynx.



**[Table/Fig-6]:** Showing a case a disseminated or 'malignant' rhinosporidiosis involving: (a) both nasal cavities; (b) conjunctiva of left eye; (c) left true vocal cord; (d) trachea; and (e) skin.



**[Table/Fig-7]:** Haematoxylin and eosin stained section of the excised mass showing numerous globular cysts of varying sizes representing sporangia at different stages of development suggestive of rhinosporidiosis.

A multivariate analysis was undertaken to evaluate any correlation between blood group, gender, duration of symptoms and bathing history with site of involvement. None of these variables were found to have any statistically significant relationship with site of involvement ( $p > 0.05$ ).

Postoperatively all patients were followed up upto 12 months. Of the 32 patients in this study, 2 patients were found to have recurrence (6.25%). This low recurrence rate may be explained by careful and complete excision of the rhinosporidial masses with basal cautery, careful counseling of patients following treatment regarding susceptible habits in rhinosporidiosis and preventive measures and regular follow-up with endoscopic examination at each visit. Of these 2 patients, one was the patient with disseminated rhinosporidiosis, a condition which is known to be associated with a high recurrence rate in view of seedling of uninvolved surrounding mucosa. The other patient gave history of bathing in pond a few months after discharge, which may be the cause for the recurrence and the patient was appropriately counseled.

## DISCUSSION

This study showed a higher prevalence of rhinosporidiosis among males. This finding agrees with those of other authors, as Arsecularatne et al., and Grover et al., also showed a higher male preponderance in their respective studies [5,9]. Probably since females have a lesser chance of animal contact and less frequent pond baths, this may account for the lower female prevalence. Some authors also opined that the effect of estrogen in females may provide some protection from the disease [10]. This study found that the age group most commonly affected was the 21 – 50 years age group, with the 31- 40 years age group having the highest predilection. Studies by Kutty et al., and Guru RK and Pradhan DK., found the maximum number of cases in the age-group 21 – 30 years [11,12]. Thus, it can be concluded that young and middle aged adults are the most commonly affected.

The males in this study were found to be farmers, daily wage labourers and factory workers, with the farmers being the most affected group. The females who were housewives in this study all gave a history of exposure to agricultural work of various kinds. In this study there was also a strong association between rhinosporidiosis and rural residential status, bathing in rivers and ponds and low socioeconomic status. Arsecularatne also found similar findings in his study, and found that persons from rural areas engaged in agricultural work and who took baths in ponds where animals were bathed also, were most at risk of developing rhinosporidiosis [5]. He also found that people taking bath in lakes or reservoirs suffer more from rhinosporidiosis of the upper respiratory tract and eye as compared to people taking bath in wells. Arsecularatne, Guru and Pradhan and Jain and Sahal also found in their respective studies a correlation between low socioeconomic status and occurrence of rhinosporidiosis [5,8,13]. The probable reason for this may be the poorer standards of hygiene among people of low socioeconomic status.

Patients in this study most commonly presented with complaints of nasal obstruction, followed by epistaxis, nasal discharge and nasal mass. Bandyopadhyay SN et al., showed similar results in their study [14]. Guru and Pradhan found most of their patients presented with epistaxis, followed by nasal discharge, nasal obstruction, sneezing and anosmia [8].

Guru and Pradhan in their study found a greater number of cases with nasal involvement than those with extra-nasal involvement [8]. Of those with extra nasal involvement, they found that nasopharyngeal, ocular, dermal and laryngeal involvement, in that order. Of those with nasal involvement, they found the most common site of attachment to be the lateral wall of nose, followed by the septum and floor, and the least to be the roof of the nose.

Author, Year	Total cases	Period of study	Country	Sites involved in order of priority	No. of disseminated cases	Age group	Population involved	Occupation	Bathing habits	SES	Recurrence rate
Ratnakar C et al., [15], 1992	139	4 years	India, Pondicherry	Nose, nasopharynx, eye	-	21-30 years	Rural	-	Ponds	-	-
Makannavar JH, Chavan SS [3], 2001	34	11.5 years	India, Karnataka	Nose, nasopharynx, eye, bone	-	11-20 years	-	Farmers, Students	-	-	-
Saha SN et al., [16], 2001	98	10 years	India, Kolkata	Nose, nasopharynx, eye, oral cavity, oropharynx, soft palate, larynx, skin, bone	-	21-30 years	Rural	-	Ponds	-	17.34%
Mohan Kameswaran et al., [17], 2005	22	2.5 years	India, Tamil Nadu	Nose, nasopharynx	-	20-40 years	-	-	-	-	27% in Group A 63% in Group B
Sudarshan V et al., [18], 2007	462	12 years	India, Raipur	Nose, nasopharynx, eye	7	21-30 years	-	-	-	-	-
Arseculeratne SN [5], 2010	143	14 years	Sri Lanka	Nose, nasopharynx, eye, penis	5	21-30 years	-	-	Reservoirs and lakes	-	37%
Saha J et al., [19], 2011	35	2 years	India, Kolkata	Nose, nasopharynx, lacrimal sac, oropharynx	-	-	-	-	-	-	5.7%
Ahmed NA et al., [20], 2013	54	3.5 years	India, Kerala	Nose, nasopharynx, eye	-	21-30 years	-	-	Ponds	-	-
Majumdar AB et al., [21], 2014	26	4 years	India, Bihar	Nose, nasopharynx	-	21-40 years	Rural	Agricultural work, outdoor manual labour	Ponds	Low	0%
Guru RK et al., [11], 2014	242	2 years	India, Sambalpur	Nose, nasopharynx, eye, skin, larynx	-	21-30 years	Rural	Agricultural work and labourers	Canals, ponds and tanks	Low	-
Sengupta S et al., [22], 2015	273	10 years	India, West Bengal	Nose, nasopharynx, eye, lips, palate, parotid duct, urethra, bone	-	21-40 years	Tribal	-	-	-	-
S Krishna Kumari et al., [23], 2015	23	3 years	India, Andhra Pradesh	Nose	-	11-30 years	Tribal	Students	Wells and ponds	-	-
Bandyopadhyay SN et al., [13], 2015	119	2 years	India, Kolkata	Nose, nasopharynx, eye, penis, skin, larynx, bone, muscle	1	11-20 years	-	-	-	-	5%
Manonmoy S et al., [24], 2015	30	3 years	India, Kerala	Nose, oropharynx	-	21-30 years	Rural	-	Ponds and rivers	-	-
Present study	32	3 years	India, Puducherry	Nose, Nasopharynx, larynx, Trachea, oropharynx, skin, eye	1	21-40 years	Rural	Farmers, housewives, students	Ponds and rivers	Low	6.25%

**[Table/Fig-8]:** Comparison of various case series of Rhinosporidiosis over the past 15 years and a previous series from Pondicherry.

Arsecularatne et al., found similar findings in their study [5]. In contrast, in this study, the commonest sites of primary attachment in the nose were found to be septum, inferior turbinate and inferior meatus in that order. Although the typical look of rhinosporidiosis is usually sufficient to arrive at a diagnosis, delayed presentation, extranasal presentation or involvement of multiple anatomically unrelated sites make the diagnosis difficult [14].

All cases of nasal and nasopharyngeal rhinosporidiosis were treated by wide endoscopic excision and cauterization of the base by diathermy. The case of disseminated rhinosporidiosis presented with a pedunculated rhinosporidial mass arising from the posterolateral wall of the trachea which was treated with the help of a special endotracheal tube designed by the anesthetist,

and using a 6mm rigid bronchoscope, the pedunculated mass was removed. Other lesions in the nose, nasopharynx and skin were excised with cauterization of the base. All specimens were sent for histopathological examination which confirmed the diagnosis [Table/Fig-7].

Fourteen other studies were reviewed and compared with the present study, 13 from the past 15 years and one previous study from Pondicherry conducted in 1992 [Table/Fig-8]. Most of the series are from various parts of India, with one large study from Sri Lanka. Apart from this study, only 3 other studies recorded cases of malignant or disseminated rhinosporidiosis, a rare manifestation of this disease which is difficult to treat and with a high recurrence rate. The vast majority of studies showed association of the disease

with the 2<sup>nd</sup> and 3<sup>rd</sup> decades of life, bathing in ponds and rivers and rural populations. The postoperative recurrence rate varies widely between the various studies, and may be attributed to differences in operative technique, instruments used for excision and number of patients in the series. An interesting note on comparison of the present study with the previous study from Pondicherry which was conducted more than two decades ago, is the drastic reduction in the number of patients of rhinosporidiosis presenting per year. This may be a consequence of a strong public health education program in this region which may have helped dissuade the new generation of the rural population from pond bathing and other high risk activities associated with this infection.

Rhinosporidiosis is an age old endemic scourge which has affected various parts of the world, most notably India and Sri Lanka. Although a large body of literature exists regarding this problem, postoperative recurrence rates continue to vary, and the effect of public health education in the eradication of this disease has not been taken into account. This study confirms the body of evidence regarding the epidemiology, treatment and prognosis of rhinosporidiosis. This study also highlights a rare case of malignant rhinosporidiosis, which is quite a rare entity and which is extremely difficult to treat. More importantly, this study highlights the effect of public health education regarding susceptible habits in the transmission of rhinosporidiosis, which has helped reduce the incidence of this disease in the previously endemic communities of Pondicherry region.

## CONCLUSION

Rhinosporidiosis is strongly associated with male gender, young and middle aged adults, agricultural occupations, rural residential status, a history of bathing in ponds and rivers and a low socioeconomic status. Patients most commonly present with complaints of nasal obstruction and epistaxis, and most commonly affects the nose, in which the inferior turbinate and septum are the most common sites of primary attachment. There appears to be no correlation between blood group, gender, duration of symptoms and bathing history with site of primary attachment. This study also shows that a strong system of public health education may help to reduce the incidence of this disease, and hence may even eventually lead to the eradication of this menace.

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