Socio-demographic Correlates of Rhinosporidiosis: A Hospital-Based Epidemiologic Study in Purulia, India

Sirshak Dutta¹ · Dibakar Haldar² · Mainak Dutta¹ · Sabyasachi Barik³ ·
Kaustuv Das Biswas⁴ · Ramanuj Sinha¹

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Abstract The present paper attempts to explore the socio-demographic profile of patients with rhinosporidiosis in an endemic area. A cross-sectional study was carried out in a tertiary-care hospital in Purulia district, India, including consecutive patients with histologically-proved rhinosporidiosis. Their socio-demographic profiles were obtained through a pre-designed proforma with given epidemiologic parameters. Data was statistically analyzed with inputs from literature review. Of the 39 patients included, 87 % were fresh/new cases. The age-group of 10–20 years was mostly involved, with multiple peaks around 50. About 82 % were from rural background, commonly involved in cattle farming and agriculture, with a universal habit of pond-bathing. There was a male preponderance; however women were being increasingly affected. Nasal cavity was the predominant site involved; nasal obstruction and epistaxis were the primary complaints. About 13 % had recurrent lesions that were statistically related to higher age-group (≥ 15 years) and occupation (agriculture, labor). Rhinosporidiosis is predominantly the disease of young rural adults engaged in field activities and habituated to pond-

bathing. A bimodal age distribution was noticed. The present article provides an update on the socio-demographic perspectives of rhinosporidiosis in an endemic zone. It also summarizes the factors that would identify the vulnerable population and help formulate preventive measures.

Keywords Rhinosporidiosis · *Rhinosporidium seeberi* · Epidemiology

Introduction

Rhinosporidiosis is a chronic granulomatous disease caused by *Rhinosporidium seeberi*—a fungus-like protozoa belonging to the class Mesomycetozoea [1]. However, there exist alternative hypotheses that suggest non-fungal etiology of the disease. These include the prokaryotic cyanobacterium, *Microcystis aeruginosa* [2, 3], and the proposed theory that *R. seeberi* is related to a group of fish parasites (the DRIP clade: Dermocystidium, rosette agent, ichthyophonous and psorospermium) [4] that flourish in the hot and humid climate. Accordingly, coastal areas of the tropics form the endemic zones. In fact, most reports of rhinosporidiosis in the world are from Sri Lanka and the coastal states of India. Definite mode(s) of transmission and the host or natural reservoirs are still unknown, but presumably, the disease gets transmitted through infected soil and stagnant water. The practice of pond-bathing is considered a potential reason for the prevalence of the disease in rural population [1]. This also explains the predilection of involvement of accessible mucosal sites, most commonly the nose and nasopharynx, where the organism gains access through traumatized epithelium [5]. Over 70 % of the reported cases were in the nasal mucosa and about 15 % in the eye [6]. The disease is common in

✉ Mainak Dutta
duttamainak@yahoo.com

¹ Department of Otorhinolaryngology and Head-Neck Surgery, Medical College and Hospital, Kolkata, 88, College Street, Kolkata, West Bengal 700073, India

² Department of Community Medicine, Bankura Sammilani Medical College and Hospital, Bankura, West Bengal, India

³ Department of Otorhinolaryngology, Debra Multi/Superspeciality Hospital, Paschim Medinipur, West Bengal, India

⁴ Department of Otorhinolaryngology and Head-Neck Surgery, Institute of Post-Graduate Medical Education and Research, Kolkata, West Bengal, India

West Bengal, one of the states in eastern India and the origin of the present study, although it is perhaps not as endemic as the coastal areas of Southern India [7]. The hot weather in most part of the year, the habit of pond-bathing due to relative scarcity of clean water in the villages, and agriculture being the primary mean of livelihood could account for the increased reporting of rhinosporidiosis from certain terrains of West Bengal [8], like the arid districts of Bankura and Purulia. In this paper we have attempted to present the epidemiologic perspectives of rhinosporidiosis in the Purulia district of West Bengal highlighting its socio-demographic correlates.

Materials and Methods

A hospital-based cross-sectional case series study was carried out for a period of 1 year (April 2013–March 2014) in the Department of Otolaryngology of Deben Mahato (Sadar) Hospital, a tertiary care health facility in Purulia, West Bengal. The study was approved by the Institutional Ethical Committee, and informed consent in written was obtained from the patients (or from their guardians, as the case may be). Consecutive patients histologically confirmed to have rhinosporidiosis were included in the study. Data were collected by interviewing the patients/guardians, examining the medical records and laboratory reports using a pre-designed and pre-tested questionnaire. Information pertaining to age, gender, residence, religion, caste, occupation, bathing habit, cattle rearing, site of involvement, disease status (i.e., fresh or recurrence) etc. were gathered. Altogether 39 patients were considered.

Statistical framework Data were analyzed using Microsoft Excel and Statistical Package for the Social Sciences (SPSS) software version 16. Various statistical methods like charts and tables were used for data display. Results were described by calculating the mean, standard deviation, median, proportion etc., and independent *t* test, Chi square/Fisher exact test, Odds ratio (OR) with 95 % confidence interval (CI) were used to explore association between the variables. *p* value of ≤ 0.05 at 5 % level of accuracy was considered as significant.

Results

Analysis of the relevant epidemiologic parameters (Table 1) revealed that the patients were predominantly male (~59 %), rural residents (~82 %), and backward class Hindus. Majority were residents of Purulia (~85 %), and the rest were referred from neighboring districts. Almost everyone (97.44 %) was reportedly habituated with pond-bathing. About 54 % belonged to the age-group of

Table 1 Distribution of patients according to some socio-demographic parameters

Attributes	Number	Percentage
<i>Gender</i>		
Male	23	58.97
Female	16	41.03
<i>Residence</i>		
Rural	32	82.05
Urban	7	17.95
<i>Religion</i>		
Hindu	31	79.49
Muslim	8	20.51
<i>Caste</i>		
General	17	43.59
Backward class	22	56.41
<i>Age group (years)</i>		
<15	18	46.15
≥ 15	21	53.85
<i>Occupation</i>		
Student/none/house wife	26	66.67
Agriculture/labor	13	33.33
<i>Cattle farming</i>		
Present	23	58.97
Absent	16	41.03
<i>Family history of contact</i>		
Present	5	12.82
Absent	34	87.18
<i>Disease status</i>		
Fresh	34	87.18
Recurrent	5	12.82
<i>Symptoms</i>		
Nasal obstruction only	15	38.46
Epistaxis only	14	35.90
Both	10	25.64
<i>Body parts involved other than nose</i>		
Yes	6	15.38
No	33	84.62
<i>Resident district</i>		
Purulia	33	84.62
Others	6	15.38

N = 39

15 years and above. One-third of the study population was engaged in agricultural activities and about 59 % in cattle farming. Nearly 87 % were fresh/new cases.

Average age was estimated to be 20.87 ± 13.32 years (median = 17; range 6–57) (Fig. 1; Table 1). Age was not found to vary across gender (mean $\pm$ SD = 20.63 ± 12.52 years vs. 21.04 ± 14.12 years, unpaired *t* = 0.095 and *p* = 0.925 at *df* 37), residence (mean $\pm$ SD = 20.06 ± 13.73 years vs. 24.57 ± 11.41 years, unpaired *t* =

Fig. 1 Age distribution of the patients. Note the multiple peaks around the age-group of 45–55 years

Table 2 Distribution of patients showing the disease status and its relation with epidemiologic parameters like age and occupation

Attributes	Disease status		Total no. (%)	<i>p</i> at <i>df</i> 1*	OR (95 %)
	Fresh no. (%)	Recurrence no. (%)			
<i>Age category (years)</i>					
<15	18 (52.94)	0	18 (46.15)	0.050	1.312 (1.033–1.667)
≥15	16 (47.06)	5 (100)	21 (53.85)		
<i>Occupation</i>					
Agriculture/labor	9 (26.47)	4 (80.0)	13 (33.33)	0.035	11.11 (1.092–113.061)
Student/none/house wife	25 (73.53)	1 (20.0)	26 (66.67)		

N = 39

* As per Fisher exact test

0.080 and $p = 0.424$ at *df* 37), and caste (mean $\pm$ SD = 20.78 ± 9.89 years vs. 20.9 ± 14.33 years, unpaired $t = 0.024$ and $p = 0.981$ at *df* 37). Most patients (53.85 %) were in the age-group of 10–20 years—the age of peak incidence. However, another small peak (15.38 %) was found around 50 years of age. Thus the present study revealed bimodality in age distribution of the patients (Fig. 1).

Analysis showed a male to female ratio of 1.44 (Table 1). Sex distribution among the patients had statistically insignificant variation across the age category [$\chi^2 = 0.063$, $p = 0.802$ at *df* 1; OR 0.848 (0.235–3.060)], caste [$p = 0.711$ as per two tailed Fisher exact test] and residential status [$p = 1.0$ as per two tailed Fisher exact test].

Nasal obstruction was the predominant symptom in 38.46 % of the subjects, closely followed by epistaxis (~36 %). About 26 % of the patients however had both the symptoms as their chief complaints (Table 1). Neither nasal obstruction nor epistaxis did show any variation across disease status, site of involvement, gender, as well as age category ($p = 1.00$ as per two tailed Fisher exact test in all cases).

Recurrence was found to be ~13 % among the presenting patients (Table 1). It was more common among the

higher age-group and those engaged in agricultural activities and labor. The difference was statistically significant (Table 2). However, recurrence was out of influence of the other factors like gender, residence, religion, caste, cattle farming, family history of rhinosporidiosis, site of affection etc. [$p > 0.05$ as per Fisher exact test].

Discussion

The clinico-histologic profile of rhinosporidiosis has been discussed many times in the current literature. It appears that, like tuberculosis, rhinosporidiosis can affect any organ system which might not necessarily be anatomically contiguous [9]. However, most reports in recent years, evident from the PubMed/MEDLINE® search with the keyword “rhinosporidiosis”, are single case descriptions at unusual, extranasal sites with varied and atypical presentations. In the last decade, barring the article by Arseculeratne et al. [10] that described the “patterns of rhinosporidiosis in Sri Lanka”, there has been no population-based or hospital-oriented study exclusively dedicated to the epidemiologic aspects of nasal rhinosporidiosis. The present article has attempted to explore the patient

characteristics using the standard epidemiologic parameters to describe rhinosporidiosis and has included subjects from one of the most highly reported areas of West Bengal.

The existing literature suggests the mean age of reporting with nasal rhinosporidiosis is predominantly the second and third decade—the optimum age of outdoor activities [8, 10]. Average age of the patients in our study was 20.87 ± 13.32 years. Evidently, rhinosporidiosis predominantly affects young adults in their most productive period of life. Although there are reports where the third decade has often been said to be the most vulnerable, some of them actually included extranasal sites [11, 12]. For example, the average age was reported as 30.42 ± 16.89 years by Mittal et al. [11] in their study at a tertiary-care hospital in South India. But it considered patients of ocular and adnexal rhinosporidiosis as well. However, one of the most interesting observations in our study that has not been reported before is the bimodal distribution of age, with multiple peaks around 50 years. Although it is understandable that a community-based study in different settings with representative and adequate sample would have been appropriate for the next level of evidence to prove this trend, our finding does provide the caveat that the middle-aged and the elderly are not immune to the disease.

Like the previously published data on gender predilection in rhinosporidiosis [5, 8, 10–13], this study has also found a male predominance. This is apparently because menfolk remain primarily engaged in outdoor activities. More than 82 % of our study population belonged to rural areas where the primary livelihood was agriculture, cattle farming and labor. However, women also took an equal and active part in these activities. The skew in contracting the disease in favor of men therefore needs further exploration and follow-up. One important revelation here is that the incidence of rhinosporidiosis in women is on the rise. The male: female ratio as we have found is 1.44, in contrast to 2 or more as reported by some previous studies [8, 14].

The nose has been found to be the predominant site of involvement. Nasal and nasopharyngeal mucosa traditionally is the preferred site of inoculation [8, 12, 13, 15], the reason is not known though [15]. Exposed mucosa with minor epithelial breach could be a possibility, which also explain rhinosporidiosis in less common sites, like oropharynx, lacrimal sac, conjunctiva, upper respiratory tract (the laryngotracheal complex) and the urethral mucosa.

Accordingly, nasal obstruction was the predominant symptom in ~38 % of patients, followed by epistaxis (~36 %). There have been inconsistent reports regarding epistaxis as the chief complaint of nasal rhinosporidiosis. For example, Saha et al. [7] in their hospital-based study reported epistaxis in only 11.22 % of patients, whereas Das

et al. [8] found it to be 80.21 %. Irrespective of the figures, epistaxis is an important presenting complaint in rhinosporidiosis as patients often seek medical help getting alarmed due to bleeding from an apparently innocuous, chronic nasal mass that was ignored for long. *From the clinicians' perspective, a reddish polypoid nasal mass with characteristic "white-dots" (the sporangia), with a history of intermittent nose-bleed, in a villager actively engaged in outdoor activities in the field or water-bodies should be considered as rhinosporidiosis until proved otherwise.* The index of suspicion should be further raised if a definite history of pond-bathing could be elicited. In fact, pond-bathing has been found to be almost a universal practice in our study.

Another important perspective to consider is recurrence, either at the primary site or in an anatomically contiguous area. Rhinosporidiosis is notorious for recurrence. It is not unusual for small foci of lesion to persist even after meticulous surgical resection, resulting in future disease. Apart from the per-operative bleeding that obscures the surgeon's view leading to possible overlooking of small residual lesions, it has been hypothesized that spillage of blood at surgery containing *R. seeberi* spores can inoculate virgin sites. This explains involvement of areas anatomically continuous with the primary site. Thus the chances of actual recurrence vary in different published studies. While recurrence in our series was nearly 13 %, Das et al. [8] and Bhandary et al. [12] found it to be 8.33 % and 22 % respectively. Comprehending the reasons for recurrences is of utmost importance because they not only add to the healthcare expenditure, but also compromise with the patients' quality of life. Analyzing the data from our study, the epidemiologic factors statistically associated with recurrence were age-group and occupation. Significantly more recurrence in the higher age-group (≥ 15 years) and among those involved in agricultural activity and as laborers re-affirmed the hypothesis that rhinosporidiosis is a water and soil-borne disease [15]. Individuals aged 15 years and above engaged in soil/water-related activities are therefore more vulnerable. Recurrence was not significantly associated with cattle rearing, although some cases of rhinosporidiosis have been described in animals such as horses, fish and cattle [12]. Although in the present series ~98 % of the patients were pond bathers and ~59 % were associated with cattle farming, absence of significant association with the disease fails to explain whether animal-to-man transmission is actually possible. Such has not yet been proven [12], and bathing in the water contaminated by diseased cattle might be the missing link.

The study has its own limitations, like the limited sample size and duration. Although the findings are suggestive and statistically valid, they could be further strengthened on evidence had a larger population been

recruited and followed up. Lack of a follow-up period could have some impact on estimating the recurrence rate in our patient cohort. However, our study is essentially cross-sectional, and the recurrences noted were as reported by the patients at their initial presentation. Also, the study was principally based on hospital data and not in the field; else more patients could have been recruited. A population-based evaluation on a larger scale can be carried out to confirm or refute our findings, and this forms one of the future scopes with this study. However, the hospital where it was conducted is a tertiary-care center that caters to a representative population with rhinosporidiosis from an endemic zone. Since every successive patient with histologically-confirmed rhinosporidiosis has been included, there could not be any bias in selection. The epidemiologic investigations subsequently carried out with the given parameters are therefore expected to provide reliable and reproducible information.

Conclusions

Rhinosporidiosis is an important existing health problem in some areas of India as well as West Bengal. Its natural history is still not fully understood. A well-planned multicenter community-based study is needed to provide and connect all the missing links for the stakeholders to formulate strategic measures for its control and prevention. The limited information available should be utilized for reducing the incidence of the disease by raising public awareness regarding its possible modes of transmission, salient symptoms, early care-seeking, and prevention. The grass-root level health-workers need to be employed for this. A recent update on the demography of the disease from an endemic region would be useful for the purpose of proper planning and aiming the target population at their habitat and livelihood. The present article provides a comprehensive socio-epidemiologic profile of the patients with rhinosporidiosis from an endemic area, which we believe would act as a functional unit of the patient characteristics at large. This could serve in future as a potential reference to initiate a larger population-based study.

Compliance with Ethical Standards

Conflict of interest The authors declare that there are no conflicts of interest associated with the manuscript preparation and its submission.

Ethical Approval All procedures performed in studies involving human participants were in accordance with the ethical standards of

the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Human and Animals Rights This article does not contain any studies with animals performed by any of the authors.

Informed Consent Informed consent was obtained from all individual participants included in the study.

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