Rhinosporidiosis: Analysis of cases presenting to a Tertiary Care Hospital in Rural Kerala

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Abstract

Background: Rhinosporidiosis was described first by Seeberi in 1900 and is caused by Rhinosporidium seeberi. It involves nasal mucosa, and is seen in both humans and animals. It is a waterborne disease endemic to the Indian subcontinent.

Objectives: To study the clinicopathological profile of Rhinosporidiosis in rural Kerala.

Methodology: The study included 30 cases of Rhinosporidiosis over a period of 2 years. All were diagnosed on a clinical basis. All subjects were treated surgically by wide excision and electrocautery, and the specimens were sent for histopathological examination. Dapsone was given to all patients with recurrence. Subjects were followed up for complications, outcome and recurrence.

Observation: The study was carried out over a period of 2 years with patients being between 7 and 70 years of age. There were 22 male patients and 8 female patients indicating a male preponderance. The main symptoms were nasal bleeding and nasal obstruction in about 75%. Nose was the commonest site involved.

Conclusion: The study reflects the endemicity of this disease in rural Kerala. High incidence is noticed in those bathing in ponds accessed by cattle, and raising health awareness among public regarding this disease would go a long way in decreasing its incidence.

Keywords: Rhinosporidiosis, Dapsone

1. Introduction

Rhinosporidiosis has been known for many years since its first description in Argentina. It is a chronic disease, with frequent recurrence after surgery. It is endemic in south Asia, notably southern India and Sri Lanka[1]. Sporadic cases are reported from North, Central and South America, South and East Africa, Japan, the Philippine Islands, Malay States, Canada, Uganda and Iran. Increased migration of persons who acquired Rhinosporidiosis in their native asian countries has resulted in the increasing incidence of this disease in non-endemic areas. Rhinosporidiosis is an infective disease as tissue lesions are always associated with the presence of the pathogen. There is no documented evidence of cross-infection between members of the same family or between animals and humans.Some data in India show a higher incidence of Rhinosporidiosis in subjects with blood group O[2]. The probable mode of infection from the natural aquatic habitat of Rhinosporidium seeberi is through traumatized epithelium, most commonly in the nasal cavity[3]. Autoinoculation is known to cause satellite lesions adjacent to granulomas especially in the upper respiratory sites[4]. Spillage of endospores from polyps during surgery is thought to be followed by autoinoculation through the adjacent epithelium. There is evidence for hematogenous spread of rhinosporidiosis to anatomically distant sites, as evidenced by the development of subcutaneous granulomas in the limbs without breach of the overlying skin[5]. The mode of regional spread, however, is controversial.

Characteristically, rhinosporidial lesions in the nasal cavity are polypoidal, granular, and red in colour due to increased vascularity. The surface contains greyish spots representing mature sporangia. Nasopharyngeal lesions are often multi-lobed with variegated appearance. Lesions on the face and trunk
are either pedunculated, or sessile on broad bases simulating verrucous warts. Rhinosporidial granulomas clinically present as ulcerated growths mimicking malignant lesions such as sarcomas and carcinomas.

A majority of cases occur in upper respiratory sites, notably the anterior nasal cavity, the inferior turbinate, septum and floor. Rhinosporidial lesions also occur in the soft palate, nasopharynx and larynx, and rarely the buccal mucosa. The disease, while being of special interest to an otorhinolaryngologist, is of interest to dermatologists and ophthalmologists as well, through the occurrence of granulomas in the skin, subcutaneous tissues and eyes. About 15% of cases of rhinosporidiosis affect the bulbar and palpebral conjunctiva. Rhinosporidiosis of the lacrimal sac and nasolacrimal duct has also been documented[7].

The definitive diagnosis of rhinosporidiosis is by histopathology, with the identification of the pathogen in its diverse stages. The etiologic agent Rhinosporidium seeberi, initially described as a protozoan by Seeberi, started to be classified as a fungus by Ashworth. However, several enigmas still persist even today regarding R. seeberi, due to the failure of attempts at sustained in vitro culture, and the failure to establish rhinosporidiosis in experimental animals. Though molecular biological analysis of the organism’s ribosomal DNA suggests it to be a cyanobacterium, like Microcystis aeruginosa[8], some consider it a mesomycetozoan[9].

Rhinosporidiosis is rarely fatal. Hemorrhage and generalized dissemination may, however, lead to death[10].

Several trials have shown that the most successful treatment, with the lowest incidence of recurrence, is surgical excision and cautery of the base of the lesions. Unfortunately, reports regarding recurrence rates are very few. Satyanarayana et al reported a recurrence rate of 11% after surgical treatment; Khan et al. followed with 22 patients of whom 18 were treated by cutting diathermy. The latter patients had no recurrences, whereas of the four cases in which the lesions were removed with forceps and snares, all had recurrent disease. Conservative treatment as a single modality is not widely used, but in one clinical trial by Nair et al. in 1979, 32 patients with nasopharyngeal rhinosporidiosis were treated post operatively with dexamethasone sodium phosphate. 71.4% patients did not show recurrence in a three-year period and none of them needed additional surgery during that period.

2. Materials and Methods

It was a prospective study carried out from 2009-2011. This series included 30 subjects with a clinical and histopathological diagnosis of rhinosporidiosis treated at Dr.SM CSI Medical College Karakonam South Kerala.

A detailed history, including age, sex, duration of symptoms, personal habits, and area of residence was recorded. Particular focus was placed on the bathing habits and work profile of the subject. Clinical examination and diagnostic nasal endoscopy were done to localize the site and extent of lesions in each case. All cases were provisionally diagnosed on a clinical basis. History of other medical or surgical illness was also recorded. All routine hematological investigations, including blood group, of each patient were also done. All subjects were treated endoscopically by wide surgical excision and electrocautery of the base of lesion under general anesthesia. Specimen was sent for histopathological examination. In extensive and recurrent cases, Dapsone 100mg daily for 2 years was given. Subjects were followed up endoscopically at 2 weeks, monthly for the next 6 months, every 2 months up to 2 years.

3. Observation & Results

A total of 30 cases were treated over a period of 3 years (January 2009- January 2011). There were 22 males (73%) and 8 females (27%) in the study (Table 1). Patients ranged from 7 years to 70 years of age, being predominantly in the 2 nd and 3 rd decade of life (Fig1).

| Sex      | 0 - 10 | 11 - 20 | 21 - 30 | 31 - 40 | 41 - 50 | 51 - 60 | 61 - 70 | Total |
|----------|--------|---------|---------|---------|---------|---------|---------|-------|
| Male     | 0      | 4       | 9       | 5       | 3       | 0       | 1       | 22    |
| Female   | 1      | 2       | 2       | 2       | 0       | 1       | 0       | 8     |
| Total    | 1      | 6       | 11      | 7       | 3       | 1       | 1       | 30    |
All patients gave a definitive history of bathing in ponds or rivers and also had contact with cattle or were cattle grazers. Nasal obstruction (77%) was the most common symptom followed by epistaxis (73%) (Table 2).

### Table 2: Symptomology of subjects

| Side          | Nasal Obstruction | Nasal Bleeding | Nasal Mass | Oral Mass | Oral Bleeding | FB Sensation Throat | Headache/Facial Pain |
|---------------|-------------------|----------------|------------|-----------|---------------|---------------------|----------------------|
| Right         | 10                | 10             | 6          | 4         | 1             | 1                   | 0                    |
| Left          | 7                 | 9              | 1          | 1         | 1             | 3                   | 1                    |
| Bilateral     | 6                 | 3              | 5          | 1         | 0             | 0                   | 0                    |
| Total         | 23                | 22             | 12         | 6         | 2             | 4                   | 1                    |
| Percentage    | 76.67             | 73.33          | 40.00      | 20.00     | 6.67          | 13.33               | 3.33                 |

The commonest site was Right side of nasal cavity (47%) and bilateral in (20%) (Table 3)

### Table 3: SIDE INVOLVED

| Side   | Frequency | Percent |
|--------|-----------|---------|
| Right  | 14        | 46.67   |
| Left   | 10        | 33.33   |
| Bilateral | 6   | 20      |
| Total  | 30        | 100     |

Of the 30 cases 60% had single attachment and 40% had multiple attachments, (Table 4)

### Table 4: Attachment of mass

| Type   | Frequency | Percent |
|--------|-----------|---------|
| Single | 18        | 60      |
| Multiple | 12    | 40      |
| Total  | 30        | 100     |

Majority of the lesions were pedunculated (about 56.67%) (Table 5)

### Table 5: Type of mass

| Type      | Frequency | Percent |
|-----------|-----------|---------|
| Pedunculated | 17   | 56.67   |
| Sessile   | 10        | 33.33   |
| Both      | 3         | 10      |
| Total     | 30        | 100     |

The commonest blood group was B+ve (33%) followed by O+ve(26%)(Table 6)

### Table 6: Blood group distribution

| Group of the Patient | Number | Percentage |
|----------------------|--------|------------|
| A+ve                 | 6      | 20         |
| O+ve                 | 8      | 26         |
| B+ve                 | 10     | 33         |
| AB+ve                | 2      | 7          |
| A-ve                 | 2      | 7          |
| B-ve                 | 2      | 7          |
Figure 2: Blood Group Distribution

4. Discussion

Rhinosporidiosis is a disease of low incidence, limited to the endemic zones in tropical and subtropical regions of India and Sri Lanka. Literature review revealed little about the possible habitat and mechanism of transmission of this pathogen. The most accepted hypotheses are pointing to a relation between contagion and patients who had contact with backwater. Some cases are described in animals such as horses, fish and cattle in general. However, animal-to-man transmission has not been proven. In our series, all subjects had a history of taking bath in ponds. It is suggested that the contact of the nasal mucosa with contaminated water inoculated through digital microtraumas is a possible method of transmission that explains the predominance of lesions in the nasal cavities. The populations of low economic status are more frequently affected, with the majority of patients coming from agricultural zones, and those taking baths in ponds.

Cases may occur between the ages of 3 to 73 years with a maximum incidence between the ages of 20 and 30 years\textsuperscript{11}. One author reported a maximum incidence between 11-20 years. Rhinosporidiosis is more common in males than females (3:1). Although the sex distribution varies in different reports\textsuperscript{12}, this may be due to the fact that males more typically have jobs related to land and backwater. We did observe a male predominance in relation to the sexes (3:1). Most of the cases observed in our series were between 21 and 30 years of age. In order to determine the relation of any possible host factor, the blood group related data of all the subjects was analyzed. The distribution of O\textsuperscript{+}, A\textsuperscript{+}, B\textsuperscript{+} and AB\textsuperscript{+} was found to be 26\%, 20\%, 33\% and 7\%, respectively. Various studies have shown it is common in patients with O group. In our study it was predominant in patient with B group.

Soft and friable nasal polyps and mucosa can be found upon examination. Grey or yellow spots, which represent the bulging sporangia through the attenuated epithelium, give a characteristic strawberry appearance. We observed a similar pattern in appearance of the lesions in our series and found it to be an important diagnostic criterion as far as rhinosporidiosis is concerned.

The most frequent site is the nasal cavity although it is found in other localizations such as the conjunctivae, oral cavity, lacrimal sac, urethra, paranasal sinuses, larynx, skin, and bone. It can affect more than one site\textsuperscript{13}. The most common nasal sites are, in decreasing order, nasal septal mucosa, inferior turbinate and nasal floor. Our findings suggest that the nasal cavity was the common site of involvement. Right side in 46.67\% and left side in 33.33\% and 20\% cases had bilateral involvement, and 60\% cases had single attachment and 40\% had multiple sites of attachment in the nasal cavity. The progression of symptoms is slow and the presenting complaints are nasal blockage, epistaxis, nasal discharge, nasal mass, and sensation of a foreign body. Clinically it presents as a polyploid mass which is friable, painless, sessile or peduncle and finely lobated with small yellow points over the mass (strawberry aspect), corresponding to mature sporangia. It bleeds on touch. Our findings revealed a similar pattern with nasal obstruction (76.67\%) being the most common presenting symptom followed by epistaxis (73.33\%), oral mass in 20\% foreign body sensation in throat (13.3\%) The diagnosis is based on clinical history, with important epidemiologic approach, detailed otorhinolaryngological examination and histopathological confirmation.

5. Conclusion

Rhinosporidiosis is a disease of dubious etiology affecting mainly farmers and is common in
tropical regions of the world. Wide local excision with cauterization of base is the main modality of treatment, with Dapsone as an adjuvant therapy in cases of recurrence and extensive diseases. Recurrence is the rule rather than the exception. Education of the population at risk should involve appropriate public health strategies and avoidance of the unhealthy practice of bathing in ponds and rivers open to animals.

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