Rhinosporidiosis – a report of two cases

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Abstract Rhinosporidiosis is primarily an infection of the nose. Though occasional involvement of other areas in the human body has been reported, it rarely presents as a disseminated disease. We describe two cases of recurrent nasopharyngeal rhinosporidiosis, one of them with cutaneous and pulmonary involvement. The clinical manifestations and the management of both the cases are discussed.

Keywords Nasopharyngeal · Disseminated · Rhinosporidiosis

Introduction

Rhinosporidiosis is a chronic granulomatous disease, endemic in south India, Sri Lanka and some parts of Africa. Few isolated cases have been reported from other parts of the world. Disseminated rhinosporidiosis is rare. Mucosal involvement of the nose, nasopharynx and soft palate is the commonest presentation. Cutaneous involvement is not very common. Involvement of other areas is uncommon. This is a report of two cases of recurrent nasopharyngeal rhinosporidiosis, one of them with cutaneous and pulmonary involvement as well.

Case reports

Case one

A 37-year-old man presented to the outpatient with gradually progressing mass in the throat of 1 year duration associated with dysphagia for solids and thickened voice. There was no history of stridor. Past history revealed that the patient had recurrent nasal mass since 7 years and was diagnosed to have rhinosporidiosis and underwent surgical excision under general anaesthesia (GA) four times earlier at different hospitals. The last surgery was done in 2005. After the last surgery, the patient was advised dapsone, which he discontinued after a month. This time, on general examination, patient had grade three clubbing and two black, oval, crust covered lesions on the anterior chest wall and abdomen. ENT examination revealed a pinkish, granular mass in the oropharynx, extending superiorly from the nasopharynx to the hypopharynx inferiorly, which used to come up into the oral cavity on gagging (Fig. 1). There was minimal bleeding on probing the mass. Anterior rhinoscopy revealed extensive synechiae in both the nasal cavities and nasal airway was significantly reduced.

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Fig. 1 Mass in the oropharynx

Fig. 3 The cutaneous lesions

Tympanic membrane was congested and bulging on both the sides. X-ray of the soft tissue neck in the lateral view confirmed the hypopharyngeal extension. Chest X-ray posteroanterior (PA) view showed multiple opacities in both the lung fields. CT scan of the thorax also revealed multiple heterogeneous lesions in both the lungs (Fig. 2). CT guided fine needle aspiration cytology (FNAC) from these lesions confirmed rhinosporidiosis. The cutaneous lesions were excised with cauterization of the base which on histopathological examination (HPE) were proved to be rhinosporidiosis (Fig. 3).

Under GA with endotracheal intubation, the excision of the mass was done (Fig. 4) and the base in the posterior pharyngeal wall of the nasopharynx was cauterized. Tracheostomy was avoided for the fear of further inoculation into the distal airway. HPE of the specimen confirmed the diagnosis of rhinosporidiosis (Fig. 5). Patient was advised bronchoscopy for the assessment of pulmonary lesions and laser excision or lobectomy if required, which he refused.

Fig. 4 The excised specimen

Fig. 2 The cavitory lesions in lung CT

Fig. 5 HPE skin rhinosporidiosis 10× showing the mature sporangia and the keratin

Patient was put on dapsone 100 mg once daily for 6 months. Nasal synechiae were released 6 months after the initial surgery. After 8 months of follow up, there was no local recurrence. The pulmonary lesions persisted, however without further progression or symptoms.

Case two

A 60-year-old man presented with mass in the throat and change in the voice of 1 year to 6 months duration. Patient had recurrent nasal obstruction due to nasal mass since 25 years and underwent excision of the rhinosporidial mass from the nose four times earlier. The first surgery was done about 25 years ago and the last one was about 7 years ago. Patient also had pulmonary tuberculosis about 2 years ago, for which he took antitubercular treatment for 6 months. The general physical examination was normal. Local examination revealed a pinkish, granular mass in the oropharynx, extending from the nasopharynx superiorly and to the hypopharynx inferiorly (Fig. 6). All the investigations were normal including chest X-ray and sputum for acid fast bacilli (AFB). Excision was done under GA with endotracheal intubation avoiding tracheostomy. Postoperatively; patient was put on dapsone, 100 mg once

Fig. 6 The clinical photograph of the second patient

daily for 6 months. No local recurrence has been seen after 8 months of follow up.

Discussion

Rhinosporidiosis, caused by *Rhinosporidium seeberi*, is known for over a hundred years. For a long time, it was thought to be a fungal disease. Herr et al. reclassified the organism as a member of protoctistan mesomycetozoa based on the ribosomal DNA [1]. Primarily, an infection

of the nose and nasopharynx, it can also affect lips, palate, epiglottis, pharynx, larynx, trachea, bronchus, ears, conjunctiva, lacrimal sac, skin, vulva, vagina, penis, scalp, tibia, femur, hand, feet and calcaneus [2].

The presumed mode of infection from the natural aquatic habitat of the organism is through the traumatized epithelium [3]. Mode of spread is by the following means [3]:

- Autoinoculation is the explanation for the occurrence of satellite lesions adjacent to granuloma especially in the upper respiratory tract and for local spread
- Hematogenous spread leads to generalized subcutaneous granulomas and involvement of viscera
- Direct inoculation of the organism in traumatized skin, otherwise called primary cutaneous type [3].

Dissemination to anatomically unrelated sites is mainly attributed to hematogenous spread [4]. Apart from these; a possibility of lymphatic spread has also been considered [5]. Involvement of tracheobronchial tree may be due to autoinoculation or hematogenous spread [6]. Spillage of endospores after the previous surgery may explain the pulmonary involvement in our case. Since there are associated cutaneous lesions over the chest wall, hematogenous spread is also possible. Cutaneous lesions could also be secondary to finger borne auto inoculation.

A diagnosis of the disease can be made by aspiration cytology and examination with 10% potassium hydroxide (KOH) or Papanicolaou smear [3]. However, a definitive diagnosis is by histopathology.

Treatment of the rhinosporidiosis is challenging. Spontaneous remission can rarely occur [5], however, treatment should be started at the earliest after the diagnosis. Diathermy excision is the treatments of choice [5]. Recurrences are still common. General opinion is that external surgery should be avoided in rhinosporidiosis for the fear of implantation [7]. Hence, tracheostomy was avoided in our cases. In spite of local diathermy excisions, recurrences are noted and new sites are getting involved. Trauma due to intubation during previous excisions may be responsible for tracheobronchial involvement. Biopsy through bronchoscopy has been recommended for such lesions [7, 8]. With the advent of CT scan and other imaging techniques, image guided FNAC is less hazardous. Recurrence and repeated diathermy excisions are acceptable in the nose and nasopharynx but in the tracheobronchial tree, this is more dangerous and requires a radical approach [7]. Now with lasers available in some centers, the choice of treatment would be excision with laser through bronchoscope. However, lasers are still not freely available in India and Sri Lanka where rhinosporidiosis is endemic and most of the patients are not able to afford the treatment because of poor socioeconomic status and poor health insurance system. Dapsone may arrest the maturation of the sporangia and promote fibrosis in stroma and can be used to prevent recurrence.

It has been observed that some unknown host factors may play a role in the development and progression of this disease [9]. Although, immunosuppression may play a role in progression of the disease, disseminated cases have been reported even in immunocompetent patients [7]. In our second case, though, the patient had pulmonary tuberculosis, the disease progression was slow compared to the first case. It is also interesting that the other family members of these two patients including their wives are not suffering from the disease though they all take bath in same pond since years. There is a possibility that antigenic variation might contribute to immune evasion [9].

Key points

- Rhinosporidiosis, though known since ages, is still not fully understood.
- Primarily a mucocutaneous disease, dissemination is rare.
- CT guided FNAC is less hazardous in the diagnosis of tracheobronchial and pulmonary rhinosporidiosis.
- Surgical excision is the treatment of choice.
- Recurrences are expected. Dapsone may reduce the chances of recurrence.
- Some unknown host factors may be responsible for the occurrence and progression of the disease.

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