

Case Report

Tracheal Rhinosporidiosis

Md. Anwar Hossain¹, Kamrul Hassan Tarafder³, Sheikh Hasanur Rahman³, Tawfiqur Rahman¹, Abdullah-AI-Mamun⁴, Md. Idris Ali¹

Abstract

Rhinosporidiosis is a chronic granulomatous infectious disease caused by rhinosporidium seeberi that usually affects the nasal mucosa and ocular conjunctiva. Involvement of trachea and bronchial tree is extremely rare. Few cases (8) have been reported in literature. We report a case of tracheal rhinosporidiosis that presented with stridor and haemoptysis with past history of surgery for recurrent rhinosporidiosis of nose and nasopharynx. Complete endoscopic excision and cauterization of base was done under general anaesthesia. No recurrent was observed for last 9 months of follow up.

Tracheo-bronchial involvement by rhinosporidiosis should be suspected when a known case of rhinosporidiosis involving upper respiratory tract presents with respiratory distress and/or haemoptysis.

Key words: Trachea, Rhinosporidiosis, Tracheostomy, Endoscopic excision, Cauterization.

Introduction

Rhinosporidiosis is a chronic granulomatous infectious disease that usually affects the nasal mucosa and ocular conjunctiva. The disease is widely prevalent in the tropics, especially in Southern India and Srilanka.¹ Rhinosporidiosis is caused by rhinosporidium seeberi a protistal microbe belonging to the

newly described class mesomycetozoa at the animal-fungal boundary.² Involvement of tracheo-bronchial tree is extremely rare and such involvement poses many diagnostic and therapeutic challenges. To the available literature review no tracheal rhinosporidiosis yet reported from Bangladesh. Here we report a case of 45 years old man with tracheal rhinosporidiosis and discuss the diagnosis and management of this rare case.

1. Medical Officer, Department of Otolaryngology Head & Neck Surgery, BSMMU, Dhaka, Bangladesh
2. Professor, Department of Otolaryngology Head & Neck Surgery, BSMMU, Dhaka, Bangladesh
3. Associate Professor, Department of Otolaryngology Head & Neck Surgery, BSMMU, Dhaka, Bangladesh
4. Consultant, Department of Otolaryngology Head & Neck Surgery, BSMMU, Dhaka, Bangladesh

Case report

A 45 year old man presented with respiratory distress for last 2 months and occasional haemoptysis for last 3 months. Severity of respiratory distress was progressively increasing in nature and patient was admitted in our department with mild to moderate degree of respiratory distress.

Correspondents: Md. Anwar Hossain, Medical Officer, Department of Otolaryngology Head & Neck Surgery, BSMMU, Dhaka, Bangladesh, E-mail: dranwar293@gmail.com

Patient was a known case of recurrent rhinosporidiosis of nose and nasopharynx and he had undergone operation in 1990, 2004

and 2010 for excision of nasopharyngeal rhinosporidiosis. His physical examination revealed a loud stridor and a bilateral respiratory wheeze. The results of ear, nose, throat examination were normal. Fibre-optic laryngoscopic examination revealed a reddish, fleshy mass attached with left antero-lateral wall of trachea with occluding the lumen of trachea. Biopsy was not done because of the

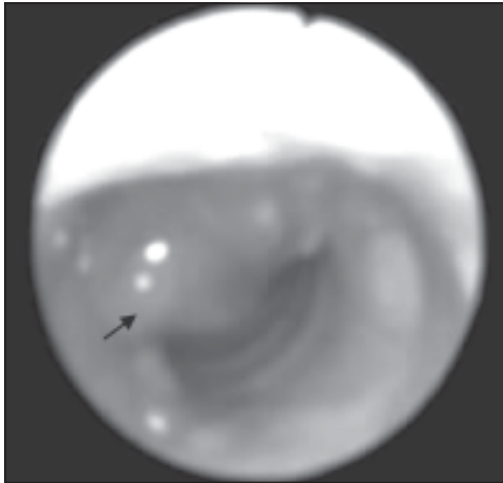


Fig-1: FOL revealed pinkish fleshy mass of rhinosporidiosis attached with tracheal wall and occluding the lumen of trachea.



Fig-2: View after complete excision and during cauterization of base of rhinosporidiosis.

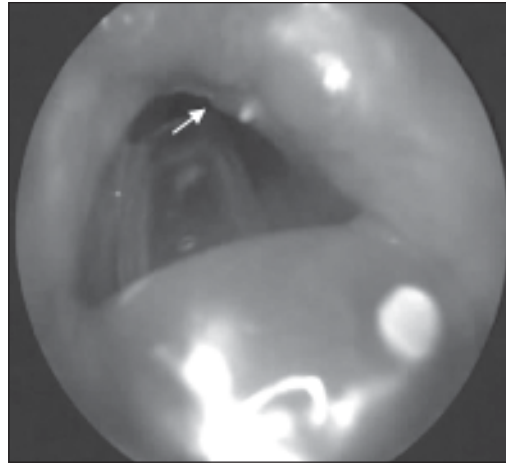


Fig-3: View after cauterization of base of rhinosporidiosis.

risk of bleeding. The patient was managed by using a combined approach of rigid endoscopy and tracheostomy which enable complete resection of the mass under direct visualization and haemostasis was achieved. The mass appeared purple red in colour and the histopathology showed many sporangia confirm the diagnosis of rhinosporidiosis. In the postoperative period we did not consider any drug therapy to prevent recurrence. Regular follow up was done to see any recurrence and no recurrence was observed within 9 months of follow up.

Discussion

Rhinosporidiosis occurs in the Americas, Europe, Africa and Asia but is most common in the tropics, with the highest prevalence in Southern India and Srilanka¹ and is caused by *rhinosporidium seeberi*. The disease results in a granulomatous inflammation of the affected tissues. The taxonomic relationship of *rhinosporidium seeberi* with other organisms remained controversial for more than a century. The organism was first described by Malbran in 1892 as a sporozoan, as a protozoan by seeber and as a phycomyces by Ashworth in 1923.³ Recently molecular

studies have shown rhinosporidium seeberi to be a protistal microbe in the newly described class mesomycetozoea at the animal fungal boundary.² Rhinosporidiosis commonly involves in nose, nasopharynx, lacrimal sac and conjunctiva in that order of frequency.⁴ It occasionally involves the lips, palate, uvula, larynx, trachea, penis, vagina and bone.⁵ Cases of disseminated cutaneous rhinosporidiosis are also reported.^{3,6} Involvement of trachea and bronchial tree is very rare. Few cases have been reported of the tracheal involvement. Rhinosporidiosis mostly affects adult men and is possibly transmitted to human subjects by means of direct contacts with spores of rhinosporidium seeberi through dust, infected clothing or fingers and through swimming in stagnant water contaminated with the spores.⁷ Tracheal rhinosporidiosis in our patient could be the result of trauma and implantation of spores during intubation for previous operations. Diagnosis of tracheo-bronchial rhinosporidiosis by means of bronchoscopic biopsy is dangerous because of the high risk of bleeding.⁴ Thomas et al. reported the bronchoscopic biopsy of bronchial rhinosporidiosis without much bleeding.⁸

The main stay of treatment for rhinosporidiosis is surgical excision by laser or electric diathermy. by using a wide tracheostomy complete endoscopic excision with base cauterization of the mass was done in our case. One study reported that rigid bronchoscopy combined with wide tracheostomy incision made the removal of tracheal rhinosporidiosis mass easier and complete.⁴ Several anti-bacterial and anti-fungal drugs have been tested clinically but the only drug which was found to have some anti-rhinosporidial effect is Dapsone which appears to arrest the maturation of the sporangia but the result are not satisfactory and the disease may recur after months or

years.^{9,10} So we did not consider any drug therapy for the prevention of recurrence in follow up period. The chance of recurrence in the case of nasal rhinosporidiosis after excision is about 10% but that of tracheo-bronchial lesion is unknown. In our case no sign of recurrence was noted in tracheo-bronchial tree with in 9 months of follow up.

Conclusion

Rhinosporidiosis in trachea could be secondary to implantation of spores from previous surgeries for nasal and nasopharyngeal rhinosporidiosis. We should inform anesthesiologist about trauma and accidental implantation of spores from nose, nasopharynx, oropharynx to lower respiratory tract by tube during endotracheal intubation. So care should be taken during endotracheal intubation. We must be careful about use of suction during surgery of rhinosporidiosis. Separate suction nostrle should be used for each nasal cavity and pharynx. A known case of rhinosporidiosis involving upper respiratory tract when presents with respiratory distress and/or haemoptysis should be kept in mind that may be involve tracheo-bronchial tree.

References

1. Fredricks DN, Jolley JA, Lepp PW, Kosek JC, Relman DA. *Rhinosporidium seeberi*, a human pathogen from a novel group of aquatic protistan parasites. *Emerg Infect Dis* 2000; 6:273-82.
2. Silva V, Pereira CN, Ajello L, Mendoza L. Molecular evidence for multiple host-specific strains in the genus *Rhinosporidium*. *J Clin Microbiol* 2005;43:1865-8.
3. Ashworth JH. On *rhinosporidium seeberi* with special reference to its sporulation and affinities. *Trans Royal Soc, Edinb* 1923; 53: 301-42.

4. Rekha P, Thomas B, Pappachan JM, Venugopal KP, Jayakumar TK, Sukumaran P. Tracheal rhinosporidiosis. *J Thorac Cardiovasc Surg* 2006; 132: 718-9.
5. Makannavar JH, Chavan SS. Rhinosporidiosis—a clinicopathological study of 34 cases. *Indian J Pathol Microbiol* 2001;44:17-21.
6. Kumari R, Laxmisha C, Thappa DM. Disseminated cutaneous rhinosporidiosis. *Dermatol Online J* 2005; 11:19.
7. Thappa DM, Venkatesan S, Sirka CS, Jaisankar TJ, Gopalkrishnan, Ratnakar C. Disseminated cutaneous rhinosporidiosis. *J Dermatol* 1998; 25: 527-32.
8. Thomas T, Gopinath N, Betts RH. Rhinosporidiosis of the bronchus. *Br J Surg.* 1956;44: 316-9.
9. Nair KK. Clinical trial of diaminodiphenylsulfone (DDS) in nasal and nasopharyngeal rhinosporidiosis. *Laryngoscope.* 1979;89: 291-5.
10. Job A, Venkateswaran S, Mathan M, Krishnaswami H, Raman R. Medical therapy of rhinosporidiosis with dapsone. *J Laryngol Otol* 1993;107: 809-12.