Tracheal rhinosporidiosis

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hinosporidiosis is a granulomatous disease that usually affects the nasal mucosa and ocular conjunctiva. The disease is widely prevalent in the tropics, especially in southern India and Sri Lanka,1 and is caused by Rhinosporidium seeberi, a protistal microbe belonging to the newly described class Mesomycetozoea at the animal-fungus boundary.2

Involvement of the tracheobronchial tree is extremely rare, and such involvement poses many diagnostic and therapeutic challenges. We report a case of tracheal rhinosporidiosis that presented with stridor.

Clinical Summary

A 48-year-old man was admitted for evaluation of breathlessness and stridor of 4 months’ duration. He had undergone operations in 1977, 1994, and 2001 for excision of nasopharyngeal rhinosporidiosis. He did not have any history of pulmonary tuberculosis, allergic diathesis, or occupational exposure to dusts or organic matter causing airway disease. Physical examination revealed a loud stridor and bilateral inspiratory wheeze. The results of ear, nose, and throat examinations were normal.

The results of his blood examination and chest radiograph were normal. Spirometry revealed flattening of the inspiratory loop of the flow-volume curve suggestive of variable extrathoracic airway obstruction. Rigid bronchoscopy showed a large, pedunculated, and fleshy mass nearly filling the lumen of the trachea above the carina. Biopsy was not done because of the risk of bleeding. A computed tomographic (CT) scan of the thorax showed the polypoidal mass (Fig 1, A and B). A virtual bronchoscopy clearly showed the degree of narrowing of the trachea (Fig 1, C) and the extent of the mass from midtrachea to 3 cm into the left main bronchus.

The patient was managed by using a combined approach of rigid bronchoscopy and tracheostomy, which enabled complete resection of the mass under direct visualization, and hemostasis was achieved. The mass appeared purple-red in color (Fig 2, A), and the histology showed many sporangia (Fig 2, B), confirming the diagnosis of rhinosporidiosis.
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 Sri Lanka, and is caused by *R. seeberi*. The disease results in a granulomatous inflammation of the affected tissues, with the nasopharynx being the most common site involved, followed by the ocular conjunctiva. The taxonomic relationship of *R. seeberi* with other organisms remained controversial for more than a century. Recently, molecular studies have shown *R. seeberi* to be a protistal microbe in the newly described class Mesomycetozoea at the animal-fungal boundary.

Rhinosporidiosis mostly affects adult men and is possibly transmitted to human subjects by means of direct contact with spores of *R. seeberi* through dust, infected clothing, or fingers and through swimming in stagnant water contaminated with the spores. Nasopharyngeal rhinosporidiosis frequently presents as a painless, friable, polypoidal growth that is pink or purple-red in color and studded with minute white dots, which are sporangia containing the spores. Nasal obstruction and bleeding are the common symptoms. Occasionally, rhinosporidiosis affects the lips, palate, uvula, maxillary antrum, epiglottis, larynx, ear, scalp, vulva, vagina, penis, rectum, and skin. Cases of disseminated cutaneous rhinosporidiosis are also reported. Involvement of the trachea and the bronchial tree is very rare.

Tracheal rhinosporidiosis in our patient could be the result of trauma and implantation of the spores during intubation for previous operations. Diagnosis of tracheobronchial rhinosporidiosis by means of bronchoscopic biopsy is dangerous because of the high risk of bleeding and the inability to visualize the full extent of the mass, especially when it obstructs the distal view, as in our case. CT imaging is the preferred investigation in such situations because it provides better details about the extent of the lesion.

Virtual bronchoscopy is emerging as a useful imaging modality for the diagnosis of airway disease, with sensitivity of about 84% and specificity of about 75% in a recent meta-analysis. It is very discriminating in the evaluation of patients with significant airway stenosis caused by a wide spectrum of pathologic conditions, especially when bronchoscopy is dangerous or might not give the full details regarding the extent of the lesion. We could avoid an open thoracotomy in our patient because we were able to determine the extent of the tracheobronchial mass lesion with virtual bronchoscopy.

Rigid bronchoscopy combined with a wide tracheostomy incision made the removal of rhinosporidial mass easier and complete. The likelihood of bleeding from the friable and highly vascular mass makes surgical intervention difficult in the case of tracheobronchial rhinosporidiosis. The chance of recurrence in the case of nasal rhinosporidiosis after excision is about 10% but that of tracheobronchial lesions is unknown. Antimicrobials cannot be recommended in routine clinical practice for the treatment or prevention of rhinosporidiosis because their efficacy is doubtful.

References