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Original Research Article

Laryngotracheal Rhinosporidiosis: A Case Study

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Abstract:

Background: Rhinosporidiosis is a chronic granuloma occurring in almost all parts of the body resulting due to infection of Rhinosporidium seeberi. Most common sites are mucous membrane of nose and nasopharynx, but also occur in other sites like oral mucosa, lips, palate, trachea, larynx, lacrimal sac, penis, scalp, skin, vulva and bone. Rhinosporidium seeberi survive in water bodies in few endemic areas with its spread mainly dominated by Natural trans-epithelial penetration. Demonstration of the organism confirmed from the pathological sites either by direct examination or by histopathological microscopy confirms the disease. Treatment consists of total excision and cauterization of its base. Among the sites mentioned Larynx is one of the rarest sites.

Aim of the Study: To present an unusual case of Rhinosporidiosis in multiple sites of Air passage successfully excised and confirmed on Histopathology.

Materials: A male aged 44 years presenting with hoarseness of voice on examination showed a polypoidal mass on the aryepiglottic fold, Left vocal cord and subglottic region. Biopsy from all the sites was done which revealed on Histopathology as Rhinosporidiosis. Results: Micro laryngeal surgery under G.A. with cuffed endotracheal tube. Visualization achieved sinus endoscope attached to Storz camera and monitor. Bleeding was present from multiple sites.

Conclusions: Laryngotracheal involvement of Rhinosporidiosis poses many diagnostic and therapeutic challenges because of the potential for bleeding and aspiration. The present case could suggest a relationship between nasopharyngeal and laryngotracheal lesions in the form of systemic dissemination or spillage of spores from the nasopharynx into the larynx during previous episodes of bleeding and surgery. More studies are required to substantiate this possibility.

Keywords: Rhinosporidiosis, Larynx, Micro laryngeal surgery, Hoarse voice, Polypoidal mass, bleeding mass.

Introduction

Rhinosporidium seeberi is a hydrophilic agent producing granulomatous lesions in the form of reddish polypoidal masses involving the nose, nasopharynx, and conjunctiva. The organism are mislead as they have features of fungi, parasites and bacteria by taxonomic classification. Later they were identified as aquatic eukaryotes based on their genetic sequencing and the nature of aquatics. [1]

The disease common in the tropics and sub tropics, termed as Rhinosporidiosis spreads through contaminant stagnant water bodies and soil. [2] Rhinosporidiosis produces, straw berry like polypoidal masses, pink to purple in colour, friable, bleeding on touch, pendulous, lesions wherever the disease is established. [3] The symptoms of Rhinosporidiosis vary depending upon the site of affection; nasopharyngeal Rhinosporidiosis presents with Rhinorrhea, epistaxis, and, ultimately nasal obstruction. [4] The clinical symptoms with history of bleeding from the site clinch the diagnosis usually. [5,6] But confirmation is by demonstration of sporangia of seeberi, or/and histopathology of the biopsied tissue. (7) Rhinosporidiosis is rarely found in European countries and Americas (but seen in South America), [8, 9 and 10]. Rhinosporidiosis is

reported in all ages but most commonly seen in the age group of 20 to 40 years. Male gender is commonly affected with Rhinosporidiosis. Both the above variables are explained by the fact that it depends upon the activity. [11]

The lesions are benign and slowly growing commonly seen in the tropical countries like India, Sri Lanka, Bangladesh and Pakistan. [12] The less common parts of the body affected with systemic dissemination are skin, larynx, trachea, genitalia, and bones, in these areas the lesions resemble soft-tissue sarcomas. [13] Usually many years elapse before the lesions occurring in the nose spreading to the less common areas by way of dissemination. [14] An unusual presentation of Rhinosporidiosis occurring in the Larynx and trachea is reported in this paper.

Materials:

A male patient aged 44 years hailing from South India agriculturist by profession came to the ENT OPD with the complaints of 1. Hoarseness of voice was since 06 months. Hoarseness was progressive in nature. Hoarseness started as a mild change in voice which later on turned to low pitched rough voice. The patient was a regular hard work labourer in the agriculture fields exposed to constant heat, contaminant soil and dust. Patient was a smoker; smoking about 15 beedies in a day. He was also misusing the voice. 2. The horse voice was associated with difficulty in breathing since 06 months. Difficulty in breathing was insidious in onset, gradual in progression. Dyspnea was on mild exertion. The patient also gave complaint of 3. Chronic irritation in his throat. In the past the patient gave history of having undergone surgeries in the nose and nasopharynx for complaints of bleeding from the nose and nasal obstruction; about 01 year ago. The patient was in the habit of bathing in the village pond which was used for common purpose of washing animals and clothes. No other member of his community, where he was living had similar complaints. There was no travel history to other places or countries by the patient. After thorough history taking videolaryngoscopy was performed after ENT clinical examination. It showed a reddish, strawberry-like pedunculated polypoidal mass with whitish spots on its surface attached to the left aryepiglottic fold, involving the glottis and subglottic region. The mass in the subglottic site was moving with respiration. The mass attached to the aryepiglottic fold was wide based and moving with the movement of the aryepiglottic fold. The patient was diagnosed provisionally as Haemangioma of supra glottis, glottis and sub glottis (multiple sites). Radiological investigation (MRI) neck was ordered to know the tissue infiltration and lower extent of the growth. MRI of the neck showed ill-defined polypoidal lesion in the glottis and infraglottic region arising from left lateral and anterior wall of glottis with irregular intra luminal surface measuring 15x16x21mm causing near total occlusion with poor separation from left arytenoids. Laboratory investigations such as complete hemogram, urinalysis, blood urea and sugar tests, chest radiography, fundoscopy and ultrasonography of the abdomen were normal. Human immunovirus and hepatitis B surface antigen serology was negative.

Result

Rhinosporidiosis is a chronic granulomatous disease that usually affects mucous membranes of the nose, nasopharynx, and ocular conjunctiva. As the polypoidal mass was occupying the aryepiglottic fold, Left vocal cord and subglottic region, it occurred that it could be a rare presentation. Biopsy was taken and sent for Histopathology examination from all the sites. Bleeding was present Multiple sites Laryngotracheal involvement poses many diagnostic and therapeutic challenges because of the potential for bleeding and aspiration. The present case could suggest a relationship between nasopharyngeal and laryngotracheal lesions in the form of systemic dissemination or spillage of spores from the nasopharynx into the larynx during previous episodes of bleeding and surgery. More studies are required to substantiate this possibility. Fig 1 showed the preoperative videolaryngoscopy of the patient. (Fig 1)

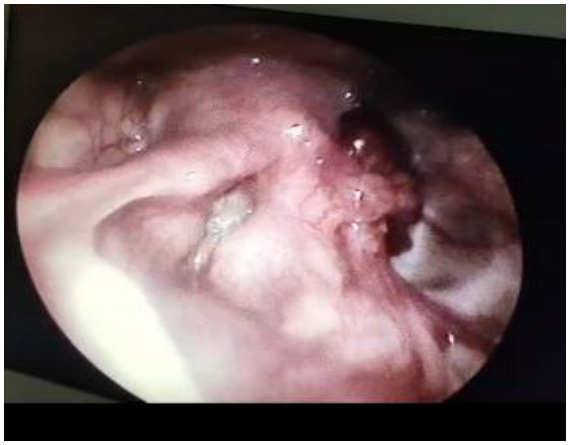

Figure 1:Pre-op videolaryngoscopy examination

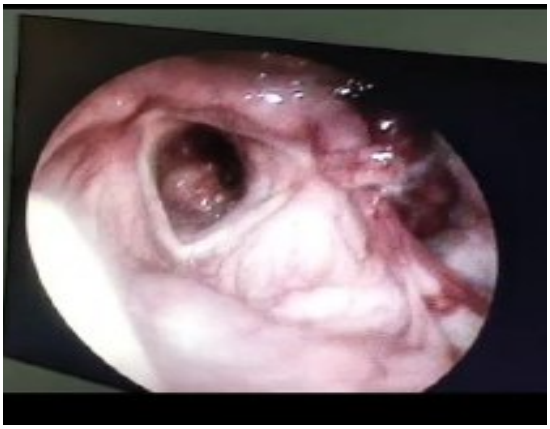

Figure 2:

- Under GA, patient was planned for Micro laryngeal surgery for wide local excision of mass.
- Growth was removed in pieces using micro laryngeal forceps from the subglottic region and also the pedunculated masses from the left aryepiglottic fold.
- Fig 2 showed the intraoperative pictures on Videolaryngoscopy. (Fig 2)

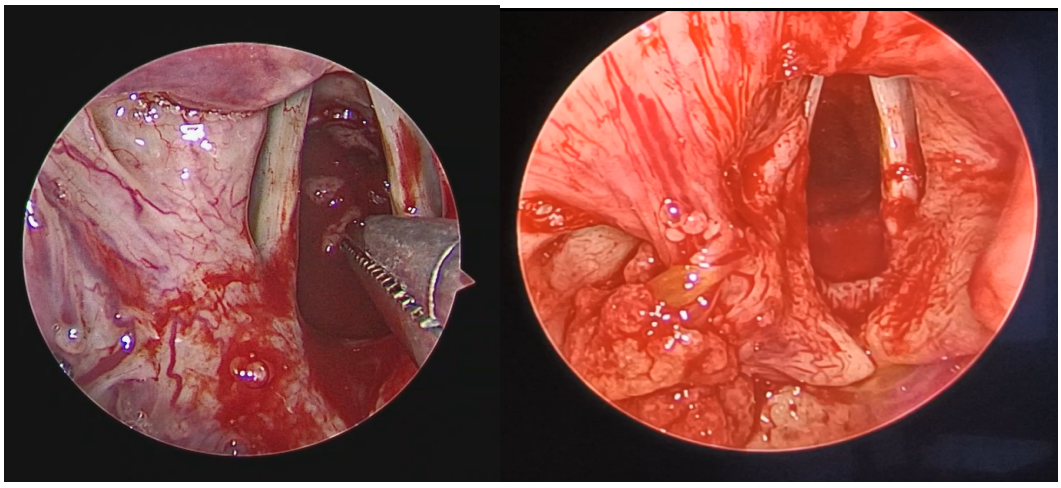

Figure 3: Intraoperative findings

Histopathology of the biopsied specimens showed

- Multiple thick walled sporangium containing endospores admixed with chronic inflammatory infiltrate of lymphocyte and plasma cells suggestive of Rhinosporidiosis.
- Sporangiums with spores were well visualized with haematoxylin and eosin stains.

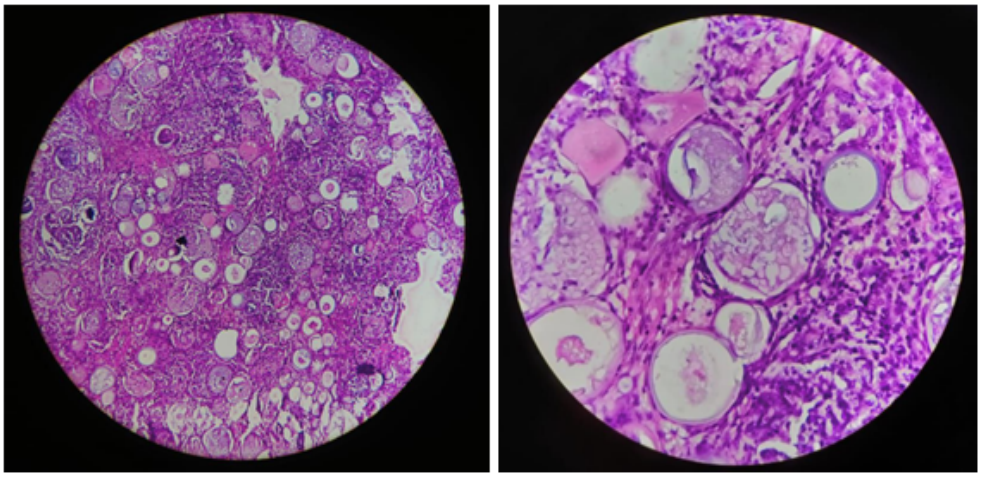

Figure 4: Histopathology

Discussion

Rhinosporidiosis was first described by O'Kinealy (1894) from eastern India. [15] The disease is caused by Rhinosporidium seeberi, a protist belonging to the newly described class mesomycetozoea at the animal-fungus boundary. [16] In 1923, Ashworth coined the term Rhinosporidium seeberi after Guillermo Seeber, who submitted a doctoral thesis in 1900. [17] Although rhinosporidiosis has been reported to occur in the Americas, Europe, Africa, and Asia, it is far more common in the tropics, and the greatest prevalence is seen in southern India and Sri Lanka. [18] Rhinosporidiosis is transmitted by direct contact with the spores of R seeberi through dust, infected clothing, or fingers, and through swimming in stagnant water contaminated with the spores. [19] Transepidermal elimination of sporangia also propagates the endospores by an active water-dependent process. In most tropical areas, rivers and lakes are the predominant media of transmission. The most prevalent site of infection is the nasal cavity. The organisms have developed several mechanisms of immune evasion (immune deviation, suppressor immune reactions, immunoglobulin binding, and antigenic variation), rendering both humoral and cell-mediated immunity ineffective. [20]

The disease affects mostly males (70 to 90%), and the incidence is greater in those aged between 20 and 40 years. Nasopharyngeal infections preferentially affect males, while ocular infection is more prevalent in females. Rhinosporidiosis produces granulomatous inflammation of the affected tissues. [21] The infection causes the development of painless intranasal papules that evolve into a large, hyperplastic, polypoidal mass that characteristically bleeds easily and has a strawberry background with white dots (representing the sporangia of the fungus). [22] The polypoidal nasal mass is usually unilateral, pedunculated, and rarely exteriorizes. Other mucosal sites involved less frequently include the palpebral conjunctiva, oropharynx, nasopharynx, maxillary antrum, larynx, external ear canal, parotid duct cyst, and genitalia. Oral and oropharyngeal lesions may produce mechanical obstruction, causing difficulty in breathing and/or food intake. Dissemination to trachea, lung, urethra, liver, genitalia, lacrimal sac, and bone is uncommon. Dissemination, which can be fatal, occurs more commonly in association with immunodeficient disorders (such as AIDS). [23] It can spread via body fluids such as ascitic fluid and blood. Airway obstruction, profuse bleeding, and multiorgan failure often precipitate death. Laryngotracheal rhinosporidiosis in our patient could have been due to implantation of spores into laryngeal and tracheal mucosa during episodes of recurrent bleeding from the nasopharyngeal lesions. [24] Bronchoscopic biopsy of the tracheal lesions could be dangerous because of the risk of bleeding and aspiration. CT of the neck and thorax is preferred in these situations to evaluate the extent of laryngotracheal lesions. Virtual bronchoscopy, a novel CT-based imaging technique that allows a noninvasive intraluminal evaluation of the tracheobronchial tree, could be useful for the diagnosis of airway disease, with sensitivity of about 84% and specificity of about 75%. [25] The histopathologic diagnosis is based on the demonstration of the characteristic thick-walled giant sporangia (measuring 60 to 450 µm or more in diameter) in various stages of development and containing sporangiospores (7 to 15 µm in diameter, and up to 12,000 in numbers). [26] R seeberi can be easily identified in hematoxylin and eosin (H&E)-stained sections. Spores and sporangia are better demonstrated by periodic acidSchiff testing, Mayer mucicarmine stain, Verhoeff-van Gieson stain, and Grocott-Gomori methamine silver stain. [27] Except for one report by Singh I, Singh A et al, [28] cultivation has not been described as successful. However, tests for assessing the viability of the organism recently

have been developed. A proportion of the spherical bodies that were provisionally regarded as the electron-dense bodies of the endospores can reduce the salt MTT (3-[4,5-dimethyl-2-thiazolyl]-2,5-diphenyl-2H-tetrazolium bromide) to a formazan that can be visualized microscopically. [29] MTT reduction as an indicator of viability of the endospores has been used to demonstrate the efficacy of several biocides. [30] Complete surgical excision is the only reliable treatment available. Cauterization/ablation of the base following surgical excision is strongly recommended as local recurrences are common due to spillage of sporangia. Rigid bronchoscopy combined with tracheostomy is useful in the removal of these laryngotracheal lesions. The likelihood of bleeding from these lesions makes surgical intervention difficult. In our case, a preliminary tracheostomy was performed followed by direct laryngoscopic excision of the laryngeal lesions. No rhinosporidial lesions were visualized in the bilateral bronchi. The recurrence rate of nasal lesions after surgery is approximately 10%, but that of laryngotracheal lesions is unknown. [31] Dapsone is claimed to be effective as it arrests the maturation of sporangia, accelerates the degenerative changes in them, accentuates the granulomatous response with fibrosis, and prevents recurrence by interfering with the folic acid metabolism of the organism. [32] Local injection of amphotericin B may be used as an adjunct treatment to surgery to prevent reinfection and spread of the disease. [33]

Conclusion

Rhinosporidiosis is a chronic granulomatous disease that usually affects mucous membranes of the nose, nasopharynx, and ocular conjunctiva. To the best of our knowledge, ours is one of the rarest reports of rhinosporidiosis, involving the larynx and trachea. Laryngotracheal involvement poses many diagnostic and therapeutic challenges because of the potential for bleeding and aspiration. Our case may suggest a relationship between nasopharyngeal and laryngotracheal lesions in the form of systemic dissemination or spillage of spores from the nasopharynx into the larynx during episodes of bleeding. More studies are required to substantiate this possibility.

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