

AN UNUSUAL ASSOCIATION OF LARYNGEAL RHINOSPORIDIOSIS WITH
SQUAMOUS CELL CARCINOMA: A CASE REPORTDr. Shristi Butta*¹, Dr. Mallika Pal² and Dr. Susmita Mukhopadhaya³¹Junior Resident (MD) Department of Pathology, NRS Medical College and Hospital.²Associate professor Department of Pathology, NRS Medical College and Hospital.³Demonstrator Department of Pathology, NRS Medical College and Hospital.

*Corresponding Author: Dr. Mallika Pal

Associate Professor, Department of Pathology, NRS Medical College and Hospital.

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ABSTRACT

Background: Rhinosporidiosis is a chronic disease caused by *Rhinosporidium seeberi* which mainly affects the nose and nasopharynx but cases have been reported in conjunctiva, skin, vulva, vagina, scalp and larynx. Larynx is comparatively a very rare site for its occurrence with only 7 cases reported till date in the world literature. Further, the association of rhinosporidiosis with squamous cell carcinoma is even rarer with very few cases reported previously. A 55year old male patient came to us with an enlarged cervical lymph node and a complaint of dysphagia with hoarseness of voice for last 6 months. **Methods:** The fine needle aspiration cytology (FNAC) was done from the enlarged cervical lymph node. 1. Chest radiography and computerized tomographic scan of the thorax. 2. Direct laryngoscopic biopsy. **Results:** FNAC was suggestive of a metastatic deposit from squamous cell carcinoma. Imaging, however, did not reveal any abnormality but as his dysphagia was getting even worse, the clinician planned a direct laryngoscopic biopsy, the histopathological examination of which yielded presence of rhinosporidiosis with few dysplastic squamous cells in the lining epithelia. Subsequent biopsy showed a histological evidence of a moderately differentiated squamous cell carcinoma along with rhinosporidiosis. **Conclusion:** This association of squamous cell carcinoma with rhinosporidiosis is rare with very few cases reported earlier. Further, laryngeal rhinosporidiosis is also a rare entity. Although clinically it presents as a reddish, polypoidal mass with a strawberry-like appearance, in cases occurring at unusual sites, histopathology remains the mainstay of diagnosis. Further, in every case of dysphagia it is very important to rule out laryngeal rhinosporidiosis as it has a potential risk of bleeding and aspiration causing life threatening complications.

KEYWORDS: rhinosporidiosis, larynx, squamous cell carcinoma.

INTRODUCTION

Rhinosporidiosis is a chronic granulomatous disease caused by *Rhinosporidium seeberi*,^[1] which is a protistan parasite^[2,3] found in aquatic bodies. This parasite was formerly classified as a fungal organism.^[2,3] The disease is endemic in India and Srilanka but cases have also been reported in East Africa and North America. The organism mainly affects the nose and nasopharynx. Extranasal involvement is rarely seen, though cases have been reported in conjunctiva, skin, vulva, vagina, scalp and larynx.^[4] Larynx is comparatively a very rare site for its occurrence with only 7 cases reported till date in the world literature. Further, the association of rhinosporidiosis with squamous cell carcinoma is even rarer with just a case documented previously.^[5]

CASE REPORT

A 55year old male smoker, farmer by profession, came to our FNAC (fine needle aspiration cytology) out door with an enlarged cervical lymph node and a complaint of

dysphagia with hoarseness of voice for last 6 months (fig 1).

FNAC was done from the enlarged cervical lymph node. The aspirate was blood mixed and particulate in nature. The smears were divided into two sets: one set was fixed in alcohol and stained with haematoxylin and eosin (H and E) whereas the other set was air dried and stained with May-Grunwald-Giemsa (MGG) stain. The smears were examined under the microscope.

The smears showed a moderate to high cellular yield. The cells were arranged in clusters and sheets with few scattered cells also noted in the background. Individual cells were pleomorphic with scanty dense cytoplasm, round to ovoid hyperchromatic nucleus, with inconspicuous nucleolus. No evidence of any keratinisation was found. The cytological diagnosis of squamous cell carcinoma, likely metastatic, was rendered (fig 2).

Two weeks later, the patient underwent a direct laryngoscopy guided biopsy. On laryngoscopic examination a proliferating growth was seen in the left vocal cord extending towards the subglottis. Biopsy was taken from the representative areas and sent for histopathological examination.

The histopathological examination showed numerous large thick walled sporangia containing numerous endospores accompanied by a mixed inflammatory infiltrate. Few dysplastic squamous cells were also seen in the lining epithelia. The histological diagnosis of rhinosporidiosis was rendered (fig 3).

Meanwhile the patient underwent a chest x ray and a computerized tomography scan of the thorax but that again was within normal limits.

The histopathological diagnosis did not collaborate with the FNAC findings. In addition, the dysphagia and hoarseness of the patient were progressing, so the clinician decided to reexamine the larynx. Similar biopsy samples were taken from the representative areas and were sent for histopathological examination. Light microscopy of the sample showed islands of moderately pleomorphic cells with scanty eosinophilic cytoplasm and round to ovoid hyperchromatic nucleus with inconspicuous nucleolus. No evidence of any keratinisation was found. In addition large sporangias laden with numerous endospores were also seen admixed with these atypical squamous islands. This was suggestive of a moderately differentiated squamous cell carcinoma with rhinosporidiosis.

The patient subsequently underwent total laryngectomy with post-operative chemotherapy for squamous cell carcinoma along with oral dapsone for preventing recurrence of rhinosporidiosis.



Fig. 1: Patient at presentation at the FNAC Outdoor.

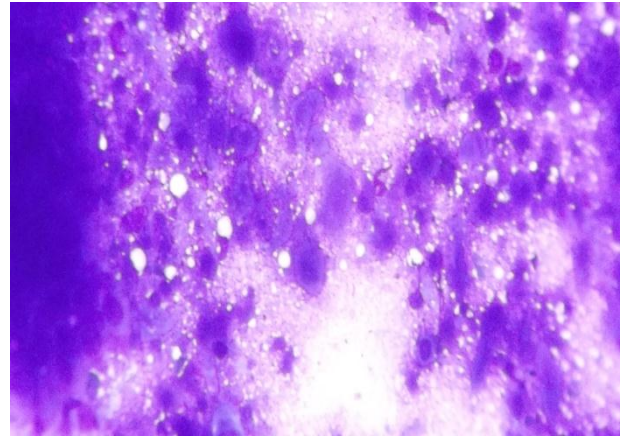


Fig. 2: FNAC of the cervical lymph node showing evidence of squamous cell carcinoma (MGG, 400X).

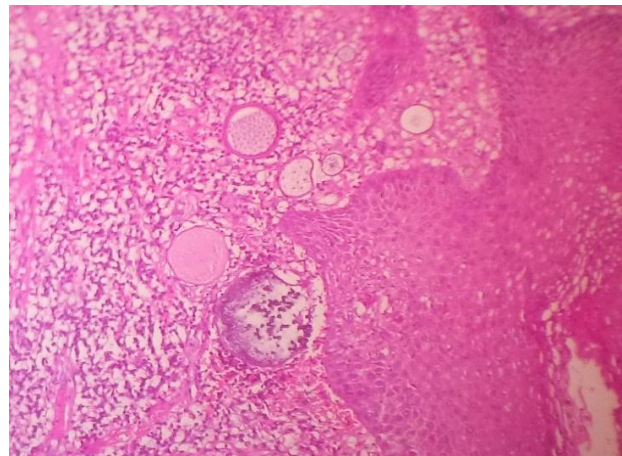


Fig. 3: Histopathology of the laryngeal biopsy specimen showing evidence of Rhinosporidiosis (H&E, 100X).

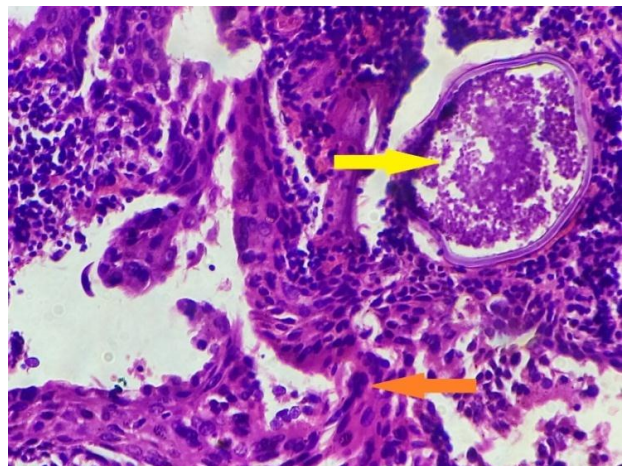


Fig. 4: Histopathology of the laryngeal biopsy specimen showing evidence of Rhinosporidiosis (yellow arrow) and islands of malignant squamous cells showing evidence of squamous cell carcinoma (red arrow), (H&E 400X).

DISCUSSION

Rhinosporidiosis, caused by *Rhinosporidium seeberi*.^[2] is an endemic disease affecting mainly the Indian subcontinent. The disease mainly affects the male population in the age group of 12 to 40 yrs. Nose and nasopharynxes are the most common sites of involvement. Though other rare sites include larynx, maxillary antrum, skin of limbs, lacrimal sac, urethra, vagina, vulva, parotid duct, bone, and rectum.^[4] Nasal disease usually presents as a unilateral nasal obstruction or epistaxis. Other symptoms may include local pruritus, coryza with sneezing, rhinorrhea, and postnasal drip. Very rarely symptoms like foreign body sensation in the throat or nose or dysphagia may be the presenting feature, like in our case.

Here in our case, besides laryngeal rhinosporidiosis the patient also had an underlying squamous cell carcinoma. This association of squamous cell carcinoma with rhinosporidiosis is even rarer with just one case documented previously by Ahluwalia et al in 1990.^[4]

Squamous cell carcinomas account for approximately 95% of all laryngeal carcinomas. The glottis is a favourable site and hence most of the patients present with hoarseness of voice. Majority of the patients also have a history of smoking and alcoholism. Progressive hoarseness of voice is the cardinal symptom of laryngeal carcinoma. Neck metastasis is more common in supraglottic cancers due to its rich lymphatic network leading to early dissemination. On the contrary glottic tumours have a poor lymphatic network. Squamous cell carcinoma often arises in a background of mucosal squamous dysplasia or carcinoma insitu. It typically presents as islands and tongues of atypical cells invading the laryngeal stroma. Other features include variable degree of individual cell keratinisation, loss of intercellular bridges and presence of keratin pearls.^[6,7]

Laryngeal rhinosporidiosis is very rare with just 7 cases reported in the world literature. It is important to diagnose laryngeal rhinosporidiosis at the earliest as this non neoplastic polyp is highly vascular and has an increased risk of bleeding and causing lethal aspiration.^[8] Further, surgical excision^[9] with cauterization of the base is the only modality of treatment. Moreover, inadequate excision can lead to a higher chance of recurrence therefore oral dapsone is given after excision to minimize the chances of recurrence^[10]

CONCLUSION

This association of squamous cell carcinoma with rhinosporidiosis is very rare with just one case reported previously by Ahluwalia et al in 1990. Further, laryngeal rhinosporidiosis is also a rare entity with just 7 cases reported in the world literature. Although clinically it presents as a reddish, polypoidal mass with a strawberry-like appearance, in cases occurring at unusual sites, histopathology remains the mainstay of diagnosis. Further, in every case of dysphagia it is very important to

rule out laryngeal rhinosporidiosis as it has a potential risk of bleeding and aspiration causing life threatening complications.

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