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Unusual Presentation Of Rhinosporidiosis - A Case Series With Review Of Literature

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Abstract

Rhinosporidiosis is a chronic infectious disease caused by the organism Rhinosporiduim seeberi . Although cases have been described worldwide, this entity is mostly found in south east asian region, affecting young people ,predominantely males of poor socioeconomic status and who are habitual of bathing in ponds, rivers, or stagnant waters. Infected soil can also be a source of infection with the high risk behaviour of nose pricking. This disease has tendency to involve nasal mucosa, however extranasal presentation are also not very uncommon. The clinical features are varied depending on the affected site, in some cases mimicking other diseases sometimes as tumor mass which can cause delay in the diagnosis. In the present case series, we have documented three case of Rhinosporidiosis with abnormal presentation , accompanied by a literature review.

Keywords: Eye Diseases, Rhinosporidiosis, Skin Diseases, Infectious, Endemic Diseases

Introduction

Rhinosporidiosis is a fungal disease which usually presents as a polypoidal growth in nasal cavity that involves anterior part of nasal septum and nasal vestibule[1]. Histopathologic examination remains one of the major diagnostic tools in mycology because it permits rapid, pre-sumptive identification of fungal infections including Rhinosporidiosis.[2] Case have been reported all over the world but it is more common in south east asia region. In India this disease is more common in the central and south region of country. In India, the disease is endemic in the states of Chhattisgarh, Kerala, Tamil Nadu, Orissa and West Bengal .In Chhattisgarh region we encountered many cases of this disease which are affecting nasal mucosa of the patients. In This study we described three rhinosporidiosis cases who presented in our hospital with very unusual presentation.

Case Series:-

Case 1:-

26 yr male patient presented with nasopharyngeal mass since 5 years , with complaint of difficulty in breathing and repeated episodes of epistaxis. Biochemical and haematological investigations were with in normal limit, On CT scan , it showed a homogenously enhancing lobulated lesion in the inferior nasal cavity, extending into the vestibule anteriorly and through the choana into the nasopharynx posteriorly . The surgical excision was done. Grossly it appears single polypoid mass measuring 11x8x5 cm. Outer surface was smooth and showed fine papillary excrescences which were fragile. Cut surface showed solid homogenous pinkish white areas. (Figure I) Histopathological examination showed polyp lined by pseudostratified columnar epithelium with plenty of lymphocyte ,

plasma cell, dilated and congested blood vessels in the submucosa. Submucosa also showed multiple sporangia (Figure II)with many endospores in different stages of development (Figure III). .No atypia or malignancy was seen. On combined clinical , radiological and histomorphological features diagnosis of nasopharngeal Rhinosporidiosis was rendered. Nasopharygeal Rhinosporidiosis with such an enlarged mass is very rare.

Case 2:-

65 year male presented with ulcerated growth affecting distal part of right sided middle finger since 1 year. Amputation was done as it was not responding to medical treatment and there was also high index of suspicion of some tumor. The growth measured 3.5x3x1.5cm. Representative sections were taken from growth as well as from resection margins. The sections showed tissue lined by Keratinized stratified squamous epithelium and Subepithelial zone showed sporangia having immature and mature spores with surrounding mixed inflammatory infiltrate. No atypia or malignancy was seen. The final diagnosis of extranasal Rhinosporidiosis was made.

Case3:-

13 year female presented with conjuctival mass since 2 years. This mass was excised and sent for histopathological examination. We received conjuctival mass measuring approximately 0.8x0.6x0.5 cm , grey white in colour. Sections showed tissue lined by non keratinising squamous epithelium alongwith subepithelial zone showing sporangia and mixed inflammatory infiltrate, congested blood vessels. No malignancy or atypia was seen. Thus the final diagnosis of Rhinosporidiosis was given to this case.

Several studies regarding rare presentation of this disease are there. Some of them are listed below :-

1.	DrPuneetaN,DrSantoshT.et al	Rare presentation of parotid duct rhinosporidiosis: A case report	Human Pathology: Case Reports Volume 14, November 2018, Pages 62-64
2	B.S. Santosh1, A. Harish Kumar1, Rajdeep Singh et al	A rare presentation of rhinosporidiosis on buccal mucosa a case report	J Oral Med Oral Surg 2022;28:5
3.	Pradeep Pradhan,Swagatika Samal.	Rhinosporidiosis of the lacrimal sac masquerading as chronic dacryocystitis: a rare presentation	Clinical Case Report and Review • Autops. CaseRep.11 • 2021(https://doi.org/10.4322/acr.2020.214)
4	Amit Kumar Dey, Rajaram Sharma, Kartik Mittal et al	Rhinosporidiosis: A Rare Cause of Proptosis and an Imaging Dilemma for Sinonasal Masses	Case Reports in Otolaryngology Volume 2016 |ArticleID 3573512 |(https://doi.org/10.1155/2016/3573512)
5.	R Chithra Barvadheesh* , B Sai Dhandapani and R King Gandhi	Soft Tissue Tumor Leg - A Rare Presentation of Cutaneous Rhinosporidiosis	Available online at www.sciencerepository.org
6	Jayakrishnan	Rare case of disseminated	*BMJCase*

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.	Kelamangalathu Narayana Kurup, Ravitheja Singasani, Simanchal P Mohanty	rhinosporidiosis with chronic osteomyelitis of the calcaneum treated by a simple technique of negative pressure wound therapy	*Reports* 2017;2017:bcr-2017221786.(http://dx.doi.org/10.1136/bcr-2017-221786)
7.	Jayanta Saha, Asim Jiban Basu, Indranil Sen , Ramanuj Sinha et al.	Atypical Presentations of Rhinosporidiosis: A Clinical Dilemma?	*Indian J Otolaryngol Head Neck Surg. 2011 Jul; 63(3): 243–246.* *Published online 2011 Jun 14. doi: 10.1007/s12070-011-0275-x*
8.	Joseph Williams, Nitin M. Nagarkar, Rupa Metha Satish Satpute and Amit Bugalia	A Rare Presentation of Rhinosporidiosis	*Indian J Otolaryngol Head Neck Surg. 2017 Dec; 69(4): 559–562*
9.	Pradeep Pradhan,Swagatika Samal	**Rhinosporidiosis of the lacrimal sac masquerading as chronic dacryocystitis: a rare presentation**	Autopsy Case Rep, vol.11, e2020214, 2021

Discussion :-

Rhinosporidiosis is a chronic granulomatous disease caused by *Rhinosporidium seeberi*, which are mostly parasites of fish, in the class Mesomycetozoa[3]. It is transmitted by contaminated water and soil as well. Risk factor is habbit of bathing in contaminated ponds . All individuals exposed to the same environment do not get infected. Spores do not enter the intact epithelium and a breach in epithelial continuity is necessary to enable infection. History of nose picking and trauma were reported in a prominent study .[4]It is primarily a disease of nose which usually presents as a reddish nasal mass which bleeds on touch. The differential diagnosis of angiomatous polyp, rhinoscleroma, angiofibroma, inverted papilloma and even malignancy should be considered in any bleeding nasal mass . Macroscopically, the typical lesion of Rhinosporidiosis is fleshy, vascular, polypoidal and granulomatous, studded with grayish white dots, present on surface of lesion .The histological features are fairly typical and the fungus may be seen in all stages of growth. In the actively growing phase the fungus shows spores,sporangia and trophozoites, while in the degenerative phase it has few viable elements in chitinous shells.The tissue response during the active phase is mainly mononuclear with some polymorphonuclear leucocytes in a vascular stroma and occasional eosinophils. In the degenerative phase there is a nongranular cell reaction with the formation of giant cells of the foreign body and Langhans types[5] The most frequent complaints are nasal obstruction and epistaxis. The nose and nasopharynx are the usual sites involved. The inferior meatus and floor of the nasal cavity are the common sites of attachment in the nose. Recurrence is very common if carefully not excised. Apart from nasal cavity the other sites of involvement are lips, palate, uvula, conjunctiva, larynx, trachea, penis, vagina and even the bone

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[6, 7]. The diagnosis is usually delayed and difficult when extranasal sites are involved

In present study we have reported three cases with unusual presentation of this disease. In first case it had produced huge mass which was around 11cm in maximum dimension and it was causing difficulty in respiration to the patient. Nasopharyngeal Rhinosporidiosis with Such a large mass is very rare. Second case is affecting nail bed of middle finger which presented as ulcerated mass. Skin involvement by this fungus is also very rare. Like wise conjuctival Rhinosporidiosis is also not very common.

With this review it is ominous that Rhinosporidiosis is having variable presentation at different site . Extranasal manifestations are also not uncommon and it can cause clinical dilemma. The most effective treatment is wide excision with a cutting diathermy and cauterization of the base of lesion with the precaution of not doing contamination of surgical site with it spores as it has increased chances of recurrence. Previously antifungal agents were also used but were ineffective [8]

Conclusion:- Nasal and extranasal rhinosporodiosis can have very unusual presentation so high index of sucspicion with relevant clinical presentation and personal history should be kept in mind as an effective surgery is the only treatment for the disease. If we are aware , we can reduce the chances of its recurrences and can provide significant relief to the patients.

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Figure's legend

Figure 1 A .Grossly it appears single polypoid mass measuring 11x8x5 cm. Outer surface was smooth and showed fine papillary excrescences which were fragile. Cut surface showed solid homogenous pinkish white areas.

Figure 1 B (scanner view 4x) polyp lined by pseudostratified columnar epithelium with plenty of lymphocyte , plasma cell, dilated and congested blood vessels in the submucosa. Submucosa also showed multiple sorangia.

Figure 1 C.(40x) Histomorphology shows characterestic sporangias with multiple spores along with with plenty of lymphocyte , plasma cell, dilated and congested blood vessels.

Figure 2 A Grossly On distal end of right middle finger shows ulcerated Growth. Cut surface shows-greyish-white area.

Figure 2B (scanner view 4x) The section shows tissue lined by squamous epithelium.Subepithelium is showing sporangia having spores with surrounding mixed inflammatory infiltrate.

Figure 2C(Low power 10x) The section shows tissue lined by hyperkeratinised stratified squamous epithelium along with sporangia. Inset is showing single sporangia having spores.

Figure 3A.(scanner view 4x) Sections show nonkeratinizing squamous epithelium with subepithelial zone showing sporangia along with

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mixed inflammatory infiltrate and congested blood vessels.

Figure 3A.(high power view 40x) Section is showing sporangia with spores.

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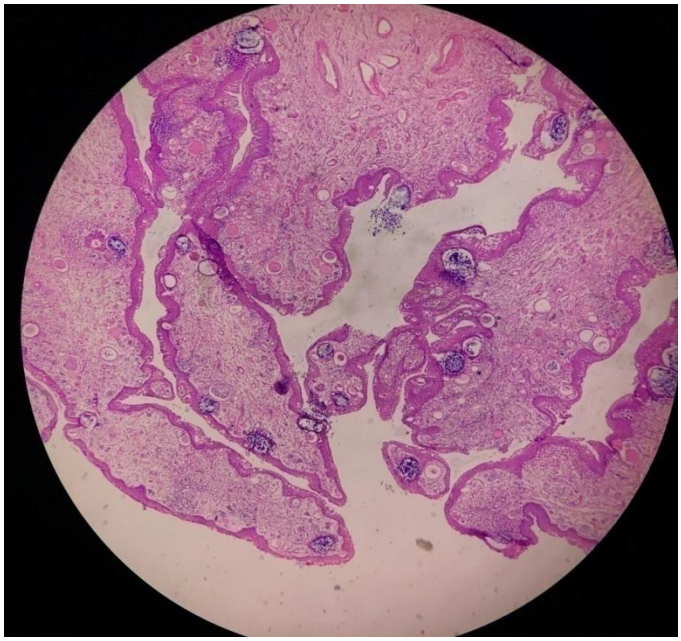

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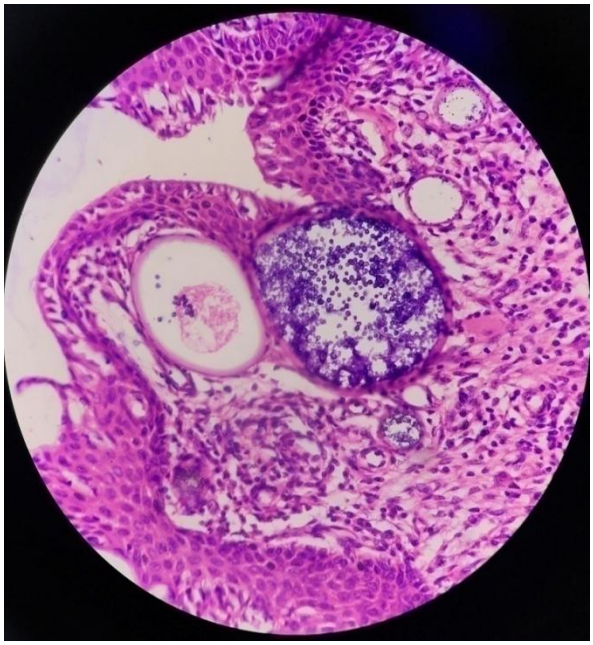

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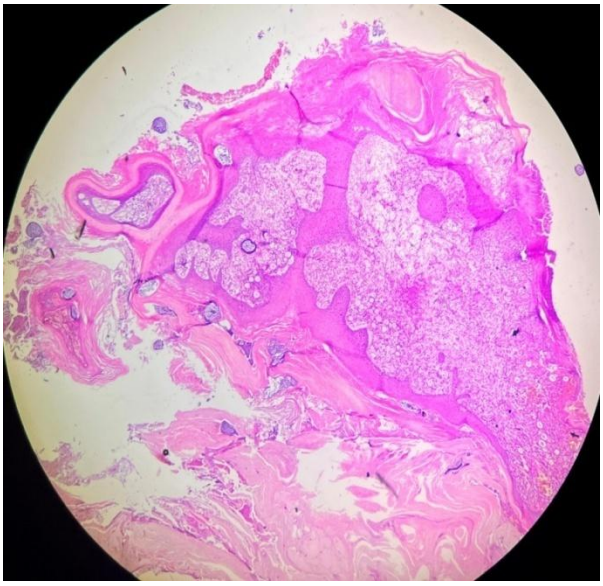

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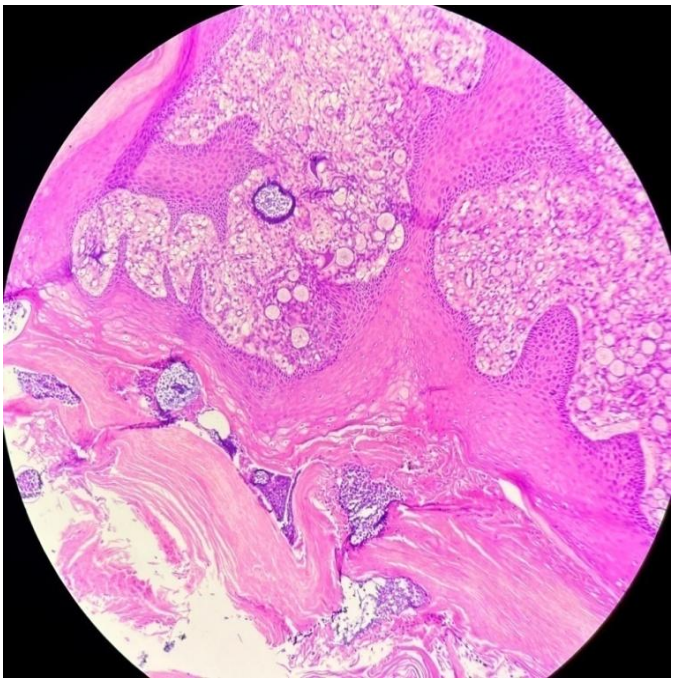

Figure 3A. (scanner view 4x) Sections show nonkeratinizing squamous epithelium with subepithelial zone showing sporangia alongwith mixed inflammatory infiltrate and congested blood vessels

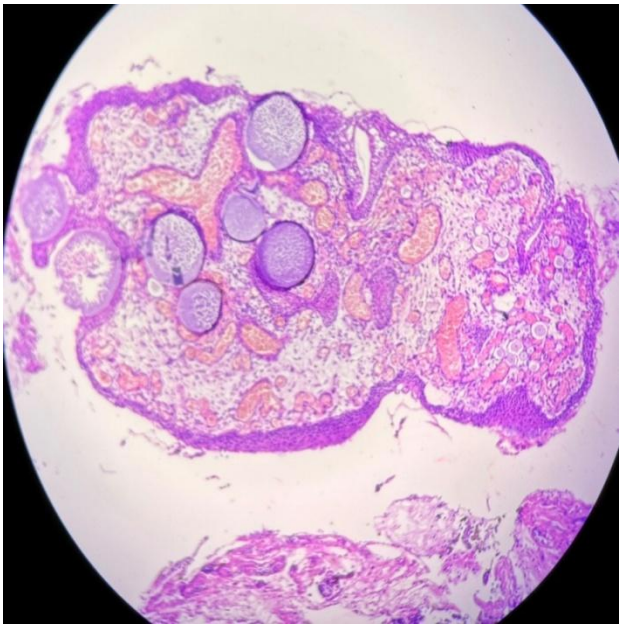

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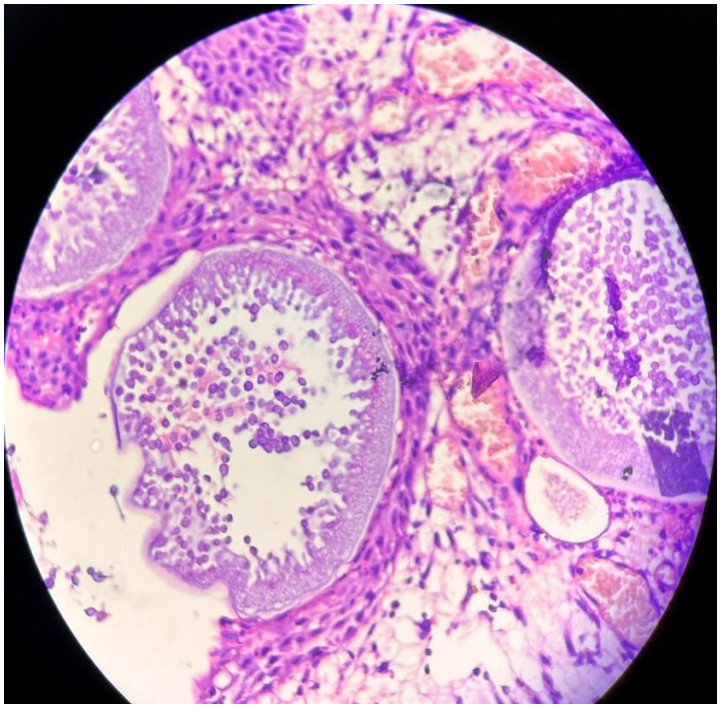
Figure 3B.(high power view 40x) section is showing sporangia with spores



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