

A Case Series of Rhinosporidiosis at Unfamiliar Sites: An Experience from a Tertiary Care Centre of Eastern India

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DOI

[10.21276/apalm.3737](https://doi.org/10.21276/apalm.3737)

Article History

Received: 17-10-2025

Revised: 04-12-2025

Accepted: 17-12-2025

Published: 05-01-2026

Abstract

Classical anatomic sites are implicated in most diseases. They help the clinician to make a substantial diagnosis. However, there exist indistinct chances of occurrence of diseases at sites that are unusual and unexpected. The same is true for the disease Rhinosporidiosis as well. We have attempted to illustrate cases of Rhinosporidiosis with presentations at infrequent sites in this compilation of cases. Tissue diagnosis has ultimately culminated in formulating the conclusive diagnosis here.

Keywords: Urethra; Conjunctiva; Cutaneous; Histopathological examination

How to cite this article

Das C, Das S, Ghatak A, et al. A Case Series of Rhinosporidiosis at Unfamiliar Sites: An Experience from a Tertiary Care Centre of Eastern India. *Ann Pathol Lab Med.* 2026;13(1):C34-C38.

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Introduction

Rhinosporidiosis is attributable to an infectious etiology. It manifests as a chronic granulomatous inflammation. The causative agent that has been isolated is *Rhinosporidium seeberi*; which has a unique status as per phylogenetic and taxonomical nomenclature. Whereas it was formerly regarded as a fungus by some, it is now considered to be an aquatic Protistan parasite

belonging to class Mesomycetozoea [1]. People belonging to the poor socio-economic strata of the society and habituated with bathing in stagnant water of ponds and lakes are commonly affected [2]. Although this disease presents as strawberry or polypoidal lesions arising out of nasal cavity, it may rarely involve other extra-nasal sites like brain, liver, genitourinary tract, gastrointestinal and hepatobiliary system as well [3].

Case Series

Materials and methods

This case series is a compilation of all the cases of Rhinosporidiosis at unfamiliar and unusual sites of occurrence that were encountered in our histopathology practice at a peripheral tertiary care centre of Eastern India within a duration of 3.5 years (January 2021 to June 2025). We have opted for Complete Enumeration method for selecting our cases. The inclusion criterion was all cases of extra-nasal rhinosporidiosis (i.e., cases of rhinosporidiosis arising anywhere in the body except from the nasal cavity). All other cases of Nasal Rhinosporidiosis have been excluded from our compilation of cases. Clinical assessment and imaging studies preceded surgical excision of the masses in the patients. Histopathological examination of post operative specimens was performed to arrive at conclusive tissue diagnosis. All the histopathological specimens underwent overnight fixation in 10% Neutral Buffered Formaldehyde. This was followed by grossing of the specimens by histopathologist and subsequently tissue processing by Automated Leica Tissue Processor. Embedding was done thereafter. This culminated in preparation of Formalin-Fixed Paraffin-Embedded (FFPE) tissue blocks. Leica Rotary microtome was used to cut sections of 4 microns thickness. Slides containing histopathological sections were stained by Hematoxylin-Eosin (H&E) stain as a routine stain and Periodic Acid Schiff (PAS) stain as a special stain.

Case report 1

A 29-year-old sexually-active male complained of post coital bleeding and a tiny pinkish nodular swelling arising out of urethral meatus. It was associated with difficulty in micturition. Urine examination was within normal limits. Initial screening for venereal disease was negative. Wedge Resection of this lesion was performed. Grossly, it was a polypoidal lesion measuring 2cmX2cmX1cm. Subsequently, multiple sporangia filled with spores were detected on histopathological examination besides dense stromal lymphocytic infiltration in the background (Figure 1). This led to the diagnosis of Urethral Rhinosporidiosis. However, there was no previous history of nasal rhinosporidiosis in this patient.

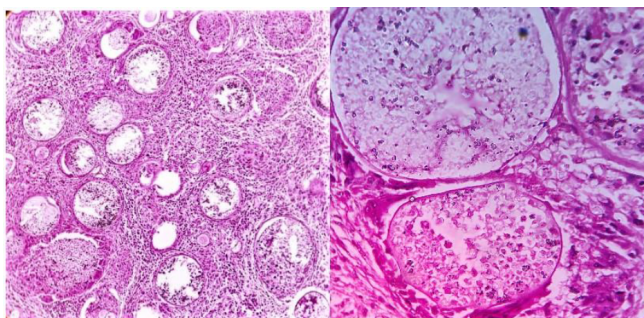


Figure 1: Histopathological section in a case of urethral rhinosporidiosis showing presence of multiple sporangia (100x; H&E); B: Sporangia are filled with numerous spores (400x; H&E).

Case report 2

A 32-year-old man was examined at the Surgery OPD for a Left arm swelling. It was a brownish, firm, rubbery, non-tender lesion and measured approximately 2.5cmX2cm. He had a past surgical history of excision of nasal rhinosporidiosis mass. It was excised and sent for histopathological examination with a clinical suspicion of common wart. However, microscopical examination of the tissue section (Figure 2) offered a dissimilar diagnosis. Presence of multiple spores bearing cystic structures were noted in the upper and middle dermis. Dense chronic inflammatory infiltrates were noted involving all layers of skin. Thereafter, this man was diagnosed to be a case of Cutaneous Rhinosporidiosis.

Case report 3

An 11-year-old girl was referred to the Ophthalmology OPD from a nearby health centre for further evaluation of an irregular mass arising from interior of her left eye. This fleshy swelling was based at Left lower palpebral conjunctiva. It had gradually increased over 8 months and caused irritation and foreign body sensation. It was meticulously excised. Histopathological

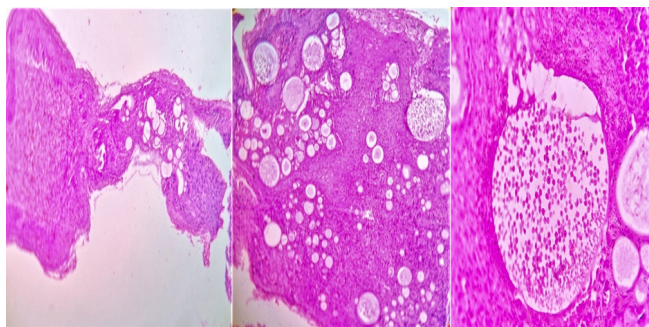


Figure 2: Cutaneous rhinosporidiosis (40x; H&E); B: Histopathological section in a case of cutaneous rhinosporidiosis showing numerous sporulated organisms (100x; H&E); C: Enlarged view of one such spore bearing structure (400x; H&E).

examination of the post-operative biopsy specimen was conducted. Presence of spores that were positively stained by PAS stain were detected just underlying the conjunctival glands and epithelium lined structures (Figure 3). A conclusive diagnosis of Conjunctival Rhinosporidiosis was hence made. Interestingly, she had never suffered from nasal rhinosporidiosis till before.

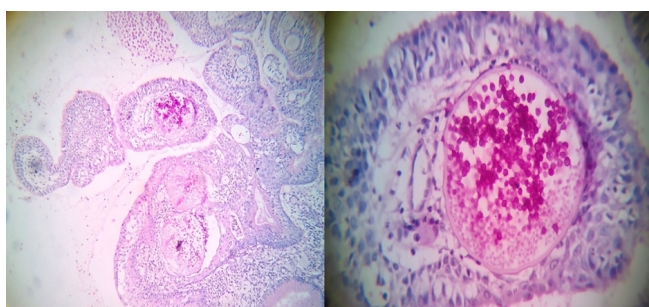


Figure 3: Histopathological section from a case of conjunctival rhinosporidiosis showing spores bearing encysted structure within conjunctival epithelium (40x; PAS); B: Enlarged view of spores in same case (400x; PAS).

Case report 4

A 43-year-old female had multiple non-tender swellings on her back, the largest measuring 5cmX4cmX2cm. They were painless, deep-seated, non-mobile and rubbery in consistency and had gradually enlarged in size over a period of 1 year. She has a past surgical history of excision for Nasal Rhinosporidiosis, 11 years ago. It was provisionally suspected to be a soft tissue neoplasm. The lady again underwent surgical excision for the swellings on her back. Histopathological examination detected presence of numerous sporulating microorganisms and various stages of their maturation (Figure 4). Dense stromal inflammation also noted. Considering the clinical profile of the patient and supplemented by histopathology findings, this was a case of Subcutaneous Rhinosporidiosis.

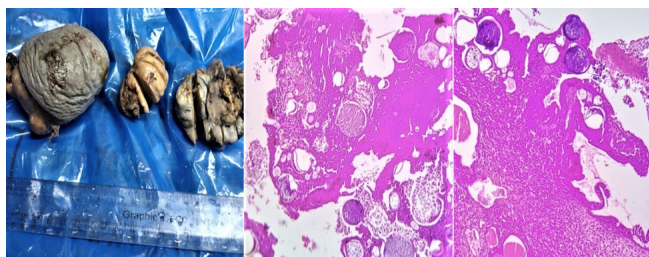


Figure 4: Gross specimen of subcutaneous rhinosporidiosis; B: Histopathological section showing numerous sporangia noted in the subcutaneous tissue (100x; H&E); C: Histopathological section showing sporangia that are densely packed with spores in a case of subcutaneous rhinosporidiosis (400x; H&E).

Case report 5

A 27-year-old lady with complaints of unilateral blocked nose and heaviness of right side of face for 4 months was attended at the ENT OPD. Per speculum examination of nose revealed an irregular, soft mass. This mass was found to be arising from Right maxillary sinus on CT scan of Paranasal Sinus. Subsequently it was excised and sent for biopsy. Grossly, an irregular greyish white soft tissue piece measuring 3x2x1cm was received. Histopathological sections showed polypoidal structure lined by pseudostratified ciliated columnar epithelium and containing numerous round sporangia filled with endospores and dense chronic inflammatory infiltrates (Figure 5). PAS stain performed on these sections yielded positive results. Thus, final

diagnosis of Sinonasal Rhinosporidiosis was made based on histopathological findings and clinico-radiological profile of the patient.

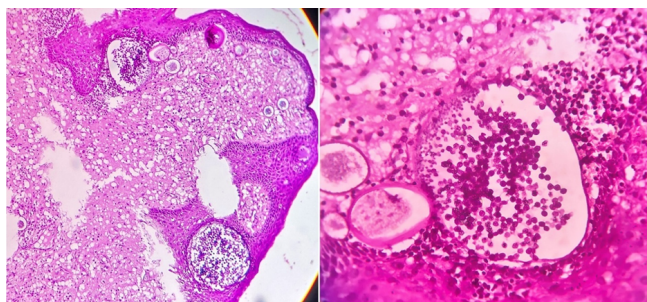


Figure 5: Histopathological section from a case of sinonasal rhinosporidiosis showing presence of sporangia in a polypoidal structure (100x; H&E); B: Enlarged view showing sporulating microorganisms in the same case. (400x; H&E).

Results

Data from all of the 5 cases of Rhinosporidiosis described above have been compiled in Table 1 as follows:

Table 1: Clinico-pathological profile of the cases.

| Sl No. | Age | Sex | Site | Clinical features | Features | History of Nasal Rhinosporidiosis | Imaging | Gross findings | Histopathological findings |
|--------|-----|-----|-----------------------|--|----------|-----------------------------------|--|---|---|
| 1. | 29 | M | Urethra | Post coital bleeding, pinkish mass from urethral meatus, Dysuria | | ABSENT | DATA NOT AVAILABLE | Polypoidal lesion measuring 2cm X 2cm X 1cm | Multiple sporangia filled with sporangiospores; Dense chronic inflammatory infiltrates |
| 2. | 32 | M | Skin | Left arm swelling | | PRESENT | DATA NOT AVAILABLE | Soft, non-tender swelling, 2.5cm X 2cm | Sporulating microorganisms in Dermis; lymphocytic infiltrates |
| 3. | 11 | F | Conjunctiva | Left eye irregular mass, foreign body sensation | | ABSENT | DATA NOT AVAILABLE | Fleshy swelling in palpebral conjunctiva | Sporangiospores containing encysted structure below conjunctival epithelium |
| 4. | 43 | F | Subcutis | Multiple deep seated, rubbery swelling over back | | PRESENT | DATA NOT AVAILABLE | Skin covered greyish white tissue piece; 5cm X 4 cm X 2cm | Numerous sporulating microorganisms in various stages of their maturation; Dense stromal inflammation |
| 5. | 27 | F | Right Maxillary Sinus | Nasal stuffiness, hemifacial heaviness | | ABSENT | CT PNS shows Right Maxillary antral mass | Irregular; greyish white tissue piece; 3cm X 2cm X 1cm polypoidal structure | polypoidal structure lined by pseudostratified ciliated columnar epithelium and containing numerous round sporangia filled with endospores and dense chronic inflammatory infiltrates |

Discussion

It is indeed intriguing to diagnose and detect cases of Rhinosporidiosis at such unfamiliar and unusual sites by histopathology; especially when there is lack of clinical suspicion of this disease entity. A study by Alam MS et al compiled cases of ocular rhinosporidiosis at sites like lacrimal sac, conjunctiva, eyelid and bony orbit [4]. A case of Urethral Rhinosporidiosis was reported by Dhakane MA et al which is similar to our case. However, our patient had remarkably normal findings on urine microscopy; whereas pyuria was noted in their case [5]. Rare case of Rhinosporidiosis involving calcaneum causing chronic osteomyelitis have been documented in scientific literature too [6]. Das C et al had conducted a similar study like ours and had reported cases of disseminated rhinosporidiosis involving muscle, bone, skin and urethra and recurrent cases as well [7]. We could also find recurrent lesions and cases involving skin and subcutis in our study. Both kinds of cases, with and without any prior history of Nasal Rhinosporidiosis were documented in our compilation of cases. However, since all of the cases were lost to follow-up, necessary clinical history of pond-bathing as a possible source of exposure to pathogen, could not be documented. We also lack in terms of data on radiological imaging findings in most of the cases.

Conclusion

Clinical examination supplemented by ancillary investigations may fail to arrive to an accurate diagnosis of Rhinosporidiosis; especially for lesions at unfamiliar and unexpected sites. Whereas, histopathological examination of excised specimen has offered the conclusive diagnosis in all of the cases enumerated above. Tissue diagnosis thus serves as gold-standard diagnosis in such clinically confusing and challenging cases. Recurrence of rhinosporidiosis is seen often and role of Dapsone has been substantially contemplated for pharmacotherapy of this disease [8]. Clinicians and pathologists alike should consider Rhinosporidiosis as one of their differential diagnoses while dealing with soft tissue lesions at varied sites.

Abbreviations: None.

Acknowledgements: None.

Informed Consent: The authors declare that written informed consent was taken from all the patients regarding the use and publication of their clinical data and/or photomicrographs, pictures of gross surgical specimens in a manner that their identity shall not be disclosed.

Funding: No funding was received for conducting this study.

Competing Interests: No conflicts of interests.

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