

#### **Case Report**

# Paediatric Nasal Rhinosporidiosis – An Overlooked Clinical Diagnosis in Non-Endemic Region

Kajol Rana', Jyotsna Madan', Abhishek Gupta², Manish Girhotra², Devajit Nath²

<sup>1</sup>Department of Pathology, <sup>2</sup>Department of Otorhinolaryngology, Post Graduate Institute of Child Health, Noida, Uttar Pradesh, India.

DOI: https://doi.org/10.24321/2454.8642.202302

# INFO

#### **Corresponding Author:**

Devajit nath, Additional Professor, Department of Pathology, Post Gradulate Institute of Child Health. **E-mail Id:** 

devajit\_nath@yahoo.co.in Orcid Id:

0000-0001-6748-8088

#### How to cite this article:

Rana K, Madan J, Gupta A, Girhotra M. Paediatric Nasal Rhinosporidiosis – An Overlooked Clinical Diagnosis in Non-Endemic Region Rec Adv Path Lab Med. 2023;9(3&4):01-04.

# A B S T R A C T

Nasal obstruction is an important clinical complaint in the adolescent age group and may lead to several early and late sinonasal complications. Rhinosporidiosis, an endemic disease of the Southern part of India, is an important differential diagnosis of nasal obstruction. Its causal organism is Rhinosporidium seeberi fungus. The condition involves the mucosa of the nose, nasal septum, middle nasal turbinate, and floor. Surgical excision of the lesion is the preferred treatment. The authors have described a case of paediatric nasal rhinosporidiosis in Northern India where the incidence is relatively low, and hence there are more chances of missing the diagnosis on clinical examination.

**Keywords:** Rhinosporidiosis, Nasal, Granulomatosis, Paediatric

Date of Submission: 2023-10-21 Date of Acceptance: 2023-11-20

# Introduction

In the paediatric age group, nasal obstruction is common and is caused by various benign and malignant conditions.<sup>1</sup> Because of the non-specific symptoms in this age group, diagnosis may be delayed which may lead to increased morbidity and mortality. Nasal masses may have a large number of differential diagnoses and a prompt pathological diagnosis is of significance because of their lymphoproliferative disorder, neoplastic nature, and the granulomatous process.<sup>2</sup>

An important, though often disregarded entity, rhinosporidiosis is a differential diagnosis for nasal granulomatosis, particularly in regions where it is prevalent, such as South America, India, and Africa. Rhinosporidium seeberi, the fungus that causes this disease, primarily affects children, teenagers, and young adults.<sup>3</sup> There are two ways that rhinosporidiosis spreads: by transepithelial penetration, and by water contamination. The fungal lesions

primarily affect the nasal mucosa of the septum, inferior nasal concha, or nasal floor.<sup>4</sup> They manifest as polyps or vascularised tumours.

The current work illustrates the case of an adolescent male with rhinosporidiosis reported in northern India where its incidence is relatively low, and hence there are more chances of missing it clinically. The final diagnosis was made after a histopathological examination.

## **Case Presentation**

A 14-year-old boy presented in ENT OPD having chief complaints of nasal obstruction for the last 6 months without any history of nasal bleeding. On anterior rhinoscopy, a nasal mass was observed in the right nasal cavity which was attached anteriorly to the floor of the nasal cavity. The mass was whitish red in colour, smooth, non-tender and did not bleed on touch (Figure 1). A differential diagnosis of a polypoidal mass was reached clinically. The mass was excised and sent for histopathological examination.

**Recent Advances in Pathology & Laboratory Medicine (ISSN: 2454-8642)** Copyright (c) 2023: Author(s). Published by Advanced Research Publications



Grossly, the mass measured 2.5 cm x 2.0 cm x 0.5 cm. The cut section was solid and greyish-white in colour. The histopathological examination on haematoxylin and eosinstained sections showed multiple fragments of respiratory epithelium with focal ulcerations and extensive squamous metaplasia. Subepithelium and underlying stroma showed multiple large thick wall sporangia filled with endospores accompanied by mixed inflammatory infiltrates in the adjacent area with few nasal mucus glands embedded in the fibrocollagenous stroma. Special stain Periodic acid-Schiff (PAS) highlighted the endospores within the sporangia (Figure 2). A final histopathological diagnosis of nasal rhinosporidiosis was reported. The patient was kept on regular monthly follow-up and showed no evidence of recurrence of the mass till now.

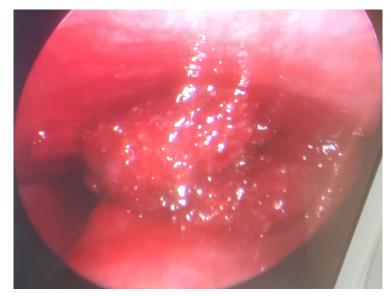


Figure 1.Nasal Endoscopy showing Red, Granular Mass in the Nasal Cavity with Yellow Pinhead Spots representing Mature Sporangia

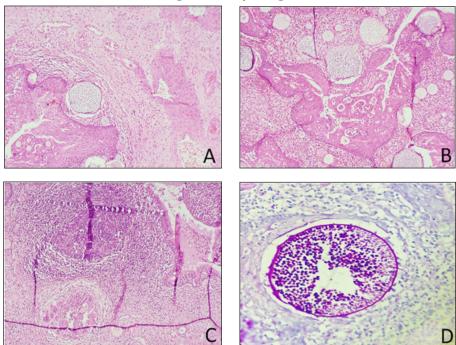


Figure 2.(A–C).Respiratory Mucosa and Metaplastic Squamous Epithelium with Underlying Mucus Gland, Dense Inflammation accompanied by Thick-walled Sporangia with Many Endospores. Adjacent Granuloma is also noted (H&E, x10). (D).Periodic Acid-Schiff (PAS) Staining Section showing Mature Sporangia with Central Mature Endospores (x10, PAS)

## Discussion

Rhinosporidium seeberi, the causative agent of rhinosporidiosis, was formerly thought to be a fungus but is now recognised as a parasite belonging to the group Mesomycetozoea—fungus.<sup>5,6</sup> Involved in the formation of steadily growing chronic granulomatous lesions, Rhinosporidium seeberi often displays benign polypoid lesions, mostly affecting the nasal, nasopharyngeal, and/ or ophthalmic regions.<sup>7</sup> With 90% of cases recorded in India and Sri Lanka and an estimated 1.4% paediatric incidence, tropical and sub-tropical areas have the highest incidence and endemicity.<sup>8</sup>

Children, teenagers, and young adults are the most frequently affected population with a male preponderance. Nasal involvement is observed in 70% of the cases, most frequently in the septal mucosa, inferior nasal concha, and nasal floor. Other locations, including the conjunctival mucosa, lacrimal sac, lungs, liver, external genitalia, and anal region, have also been documented to be involved in the literature.<sup>9</sup>

In most of the anatomical sites, lesions resemble polypoid, pedunculated or vascular masses with irregular surfaces. Prominent clinical features in case of nasal/ sinonasal involvement are those of nasal bleeding, congestion and mucopurulent rhinorrhoea.<sup>10</sup>

Rhinosporidium seeberi appear as huge, spherical structures in microscopy that range in size from 50 to 100 mm and resemble yellowish tiny spots. In addition, they highlight eosinophilic walls enclosing smaller spherical structures containing amorphous eosinophilic material (Figure 2).

The mucicarmine, periodic acid-Schiff, and Grocott-Gomori's methenamine silver stains are used to visualise the microscopic characteristics of this organism.<sup>11</sup> Although Rhinosporidium seeberi's sporangia and endospores are bigger than spherules, it shares a very similar shape with Coccidioides.

The best course of treatment for rhinosporidiosis is surgical removal, which also helps in the disease's histological diagnosis. The literature also discusses medical therapy, such as dapsone, but none of these yields results that are as good as surgery.<sup>12</sup>

Although the prognosis for rhinosporidiosis is generally favourable, clinical data indicate that timely diagnosis, active therapy, and long-term patient follow-up are necessary to detect relapses and prevent recurrences.<sup>13</sup>

#### Conclusion

Physicians should be alert regarding this uncommon entity which despite having a low incidence in the nonendemic region in the paediatric age group may present as a differential diagnosis with nasal mass, thereby making histopathological diagnosis and proper surgical management necessary.

## Acknowledgement

The authors extend their gratitude to the Institute and Technical Support Staff, Mr Utkarsh Pal, Mr Mahesh Kumar, Mr Manish Kumar, and Mr Mayank Pandey.

## Conflict of Interest: Nil

#### References

- Leboulanger N. Nasal obstruction in children. Eur Ann Otorhinolaryngol Head Neck Dis. 2016 Jun;133(3):183-6. [PubMed] [Google Scholar]
- Nwawka OK, Nadgir R, Fujita A, Sakai O. Granulomatous disease in the head and neck: developing a differential diagnosis. Radiographics. 2014;34(5):1240-56. [PubMed] [Google Scholar]
- Almeida FA, Feitoza LM, Pinho JD, Mello GC, Lages JS, Silva FF, Silva RR, Silva GE. Rhinosporidiosis: the largest case series in Brazil. Rev Soc Bras Med Trop [Internet].
  2016 [cited 2023 Sep 15];49(4):473-6. Available from: https://pubmed.ncbi.nlm.nih.gov/27598634/ [PubMed] [Google Scholar]
- Izimukwiye AI, Mbarushimana D, Ndayisaba MC, Bigirimana V, Rugwizangoga B, Laga AC. Cluster of nasal rhinosporidiosis, Eastern Province, Rwanda. Emerg Infect Dis. 2019 Sep;25(9):1727-9. [PubMed] [Google Scholar]
- 5. Ahluwalia KB. Causative agent of rhinosporidiosis. J Clin Microbiol. 2001 Jan;39(1):413-5. [Google Scholar]
- Mendoza L, Taylor JW, Ajello L. The class Mesomycetozoea: a heterogeneous group of microorganisms at the animal-fungal boundary. Annu Rev Microbiol. 2002;56:315-44. [PubMed] [Google Scholar]
- Das S, Kashyap B, Barua M, Gupta N, Saha R, Vaid L, Banka A. Nasal rhinosporidiosis in humans: new interpretations and a review of the literature of this enigmatic disease. Med Mycol [Internet]. 2011 Apr [cited 2019 Oct 21];49(3):311-5. Available from: https:// academic.oup.com/mmy/article/49/3/311/1095671/ [PubMed] [Google Scholar]
- 8. Branscomb R. Rhinosporidiosis update. Lab Med. 2002 Aug 1;33(8):631-3. [Google Scholar]
- Vélez A, Jiménez G, Hidrón A, Talero S, Agudelo CA. Rhinosporidiosis in Colombia: case series and literature review. Trop Doct. 2018;48(4):289-93. [PubMed] [Google Scholar]
- 10. Devaraja K, Sagar P, Singh CA, Kumar R. Nondisseminated rhinosporidiosis with multisite involvement in the head

and neck. Ear Nose Throat J [Internet]. 2018 [cited 2023 Sep 15];97(9):E15-7. Available from: https:// pubmed.ncbi.nlm.nih.gov/30273436/ [PubMed] [Google Scholar]

- Arseculeratne SN, Atapattu DN, Wickramaratne K. Nature and significance of the electron-dense bodies of the endospores of rhinosporidium seeberi: their reactions with MTT (3-[4,5-dimethyl-2-thiazolyl]-2,5-diphenyl-2H-tetrazolium bromide) and TMRE (tetramethyl-rhodamine ethyl ester). Med Mycol [Internet]. 2005 May [cited 2023 Sep 15];43(3):261-73. Available from: https://pubmed.ncbi.nlm.nih. gov/16010853/ [PubMed] [Google Scholar]
- Bakshi SS. Rhinosporidiosis. J Allergy Clin Immunol Pract [Internet]. 2017 [cited 2023 Sep 15];5(6):1739. Available from: https://pubmed.ncbi.nlm.nih.gov/28757369/ [PubMed] [Google Scholar]
- Chen L, Buonocore D, Wang B, Tabaee A. Delayed recurrence of sinonasal rhinosporidiosis. Am J Otolaryngol [Internet]. 2015 [cited 2023 Sep 15];36(6):778-80. Available from: https://pubmed. ncbi.nlm.nih.gov/26545470 [PubMed] [Google Scholar]