# Nasal rhinosporidiosis masquerading as pyogenic granuloma in a paediatric patient at a Zonal Referral Hospital in Central Tanzania: Case report

Zephania Abraham[1], Francis Zerd[2], and Aveline Kahinga[3]

[1]University of Dodoma College of Health and Allied Sciences
[2]Benjamin Mkapa Hospital
[3]Muhimbili University of Health and Allied Sciences School of Medicine

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**Abstract**

Being rare, rhinosporidiosis is a chronic granulomatous disease that is characterized by polypoidal lesions of the mucous membrane and is caused by Rhinosporidium seeberi. The disease commonly affects the mucous membrane of the nasopharynx, conjunctiva and palate and its very rare in our geographical location.

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**ABSTRACT**

Being rare, rhinosporidiosis is a chronic granulomatous disease that is characterized by polypoidal lesions of the mucous membrane and is caused by Rhinosporidium seeberi. The disease commonly affects the mucous membrane of the nasopharynx, conjunctiva and palate and its very rare in our geographical location.

**KEYWORDS:** Nasal mass, Rhinosporidiosis, Paediatric patient, Granulomatous

**KEY CLINICAL MESSAGE**

Nasal rhinosporidiosis lesions may greatly mimic other ordinary nasal polyps and it's crucial for otorhinolaryngologists to consider rhinosporidiosis as a differential diagnosis whenever encountering patients with nasal masses during routine clinical practice

## Introduction

Nasal rhinosporidiosis is a chronic granulomatous disease of the nose caused by Rhinosporidium seeberi.[1-5] The disease predominantly affects the mucous membrane of the nose, nasopharynx and conjunctiva**.** Rhinosporidiosis tend to occur in human beings and in animals and is more common in hot tropical climates though highly endemic in India and Sri Lanka.[6-9] Sporadic pattern for the disease has been reported in other parts of the world such as Argentina, Brazil and Africa.[9]

In terms of sex predilection, there is no racial predominance and males are ore affected than females especially those aged 15-40 years with male to female ratio being 4:1.[2,10,11]Rhinosporidiosis may be transmitted by direct contact with spores through dust, infected clothing and swimming in stagnant water.[7,8,12,13] Though the disease remains to be very rare in our country yet there are some countries that have reported several cases including Nigeria.[8] The diagnosis of nasal rhinosporidiosis is established by observing the characteristics of

the causative organisms in nasal tissue biopsies like sporangia that may be at variable stages of maturation. Since it presents like a polypoidal mass in the nasal cavity, it mimicks other diseases presenting with nasal masses [5,14-17] thus a high index of suspicion by clinicians is of importance in management of patients with nasal masses in the era of this emerging disease entity. The main stay of treatment is surgical excision of the nasal mass though a high recurrence rate has been reported.[4,5,15,17] It may lead to death immunocompromised patients.[18] To the best of our knowledge, this is the first reported case of nasal rhinosporidiosis in Central Tanzania and the 16$^{th}$ case countrywide

We are therefore reporting a case of nasal rhinosporidiosis that was managed by endoscopic surgical excision of the nasal mass and kept on oral dapsone for 6 months postoperatively.

**Case presentation**

We are presenting an 11-year old male boy who presented at our outpatient otorhinolaryngology clinic at Benjamin Mkapa Hospital with a 2-years history of right-sided nasal obstruction and intermittent epistaxis for 2 years. He had no history of cheek pain, numbness of the cheek, loss of teeth, loosening of teeth or alveolar ridge fullness. No ophthalmological, otological or neurological complaints were reported upon enquiry.

On physical examination, the patient was found to have a right-sided friable nasal mass filling the entire nasal cavity and had no nasal deformity externally. He was not pale and had no palpable peripheral lymph nodes. Ophthalmological, otological and neurological evaluation revealed normal findings. Laboratory results showed hemoglobin 12g/dl and elevated ESR (40mm/hour). A provisional diagnosis of pyogenic granuloma was established and the patient was sent for endoscopic surgical nasal mass excision under general anaesthesia and the excised specimen was sent for histopathology. Histopathological analysis revealed thick-walled sporangium containing numerous endospores (daughter spores) (Figure 1) and with further sections showing thick walled sporangium with endospores in different stages of development accompanied by a mixed inflammatory cells mainly plasma cells and lymphocytes (Figure 2).

**Figure 1:** Intermediate power view of thick-walled sporangium containing numerous endospores (daughter spores)

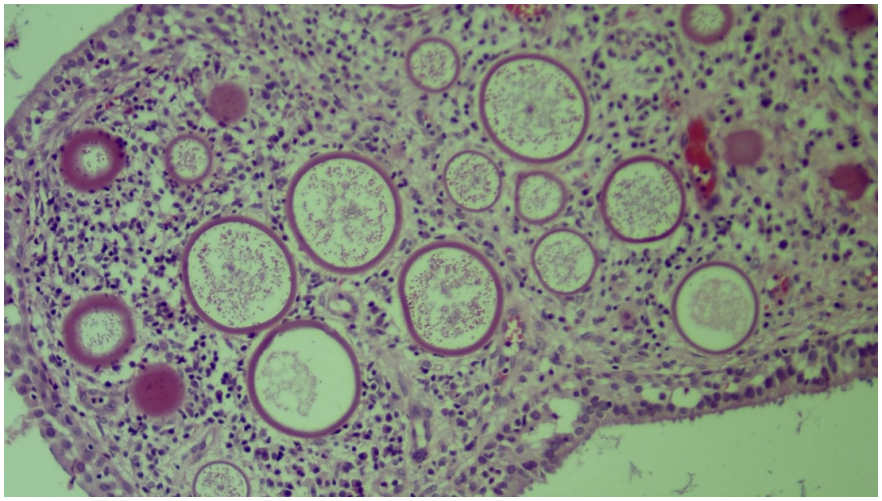
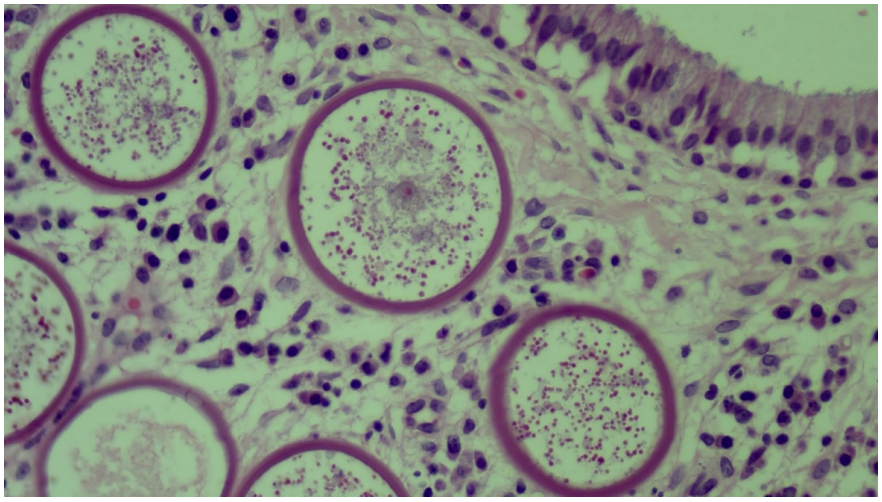


**Figure 2:** A high-power view of Large, thick walled sporangium with endospores in different stages of development accompanied by a mixed inflammatory cells mainly plasma cells and lymphocytes.



The patient was kept on oral dapsone 50mg/day for 6-months and with no residual disease recurrence after 6-months of follow up.

**Discussion**

This case report documents a case of nasal rhinosporidiosis in a paediatric patient at a Zonal Referral Hospital in Central Tanzania and so far the first documented case in Central Tanzania and the 16$^{th}$ case countrywide. The disease has been reported from about 70 countries with diverse geographical features.[7] The infrequently, isolated cases are reported in other parts of the world and are mainly due to migration.[2,9]

The disease is more common in younger age groups from the available literatures and being more common in men than women with male to female ratio being 4:1[2,10,11] These observations appear similar to what has been observed in our case report where the affected patient was an 11-year old male child.

Rhinosporidiosis and its causative organism Rhinosporidium seeberi have been known for over a hundred years and it's a rare infective chronic granulomatous disease that remains to be endemic in some part of Asia (India), although sporadic cases have been reported in America, Europe and Africa[4,5,15-17]

To date the causative organism has never been isolated in vitro, and its taxonomic position is unclear.[19]

In contrast with more recent fungal infections, some aspects of the taxonomy, morphology, ontogenesis and epidemiology of those caused by Rhinosporidium seeberi remain controversial and have not been resolved. Though now related to a group of fish parasites referred to as the DRIP clade, most pathologists and microbiologists initially considered it to be fungus on the basis of its property to be stained by fungal stains such as Gomori methenamine silver (GMS) and periodic acid-Schiff (PAS).[20,21]

Available literatures indicated that a combination of host specificities and resistance of Rhinosporidium to grow in culture may account for the failure to produce experimental rhinosporidiosis.[21]Some authors proposed the class of Rhinosporidium seeberi to be Mesomycetozoa.[13,20,21]. Fluorescent in-situ-hybridization techniques provide evidence that the natural habitat for Rhinosporidium seeberi are water reservoirs and perhaps soil contaminated by waste. In addition, other aquatic microorganisms might be relevant to a possible synergistic action in the establishment of natural rhinosporidiosis.[21]

The class Mesomycetozoa has two orders, which are the Dermocystida and the Ichthyophonida. In the order Dermocystida is the family Rhinosporideaceae that includes Rhinosporidium seeberi, Dermocystidium spp. and the rosette agent.[21]

The route of transmission for Rhinosporidium remains to be unclear even though the presumed mode of infection from the natural aquatic habitat of Rhinosporidium seeberi is through a traumatized epithelium

commonly called trans epithelial infection and this is most common in nasal sites. For Rhinosporidium seeberi, various modes of spread have been documented including; auto-inoculation through spillage of endospores from polyps after trauma or surgery, haematogenous spread to distant sites, lymphatic spread, and sexual transmission.[3,21] Rhinosporidiosis is prevalent in rural settings, particularly among people working or in contact with contaminated soil, stagnant water (ponds, or lakes) or sand.[21] In our case report, the patient gave a history of contact with contaminated pond water and was residing in a rural area. Similarly, the patient reported a history of contact with feces of infected livestock and used to work in contaminated agricultural fields. Such risk factors have been reported in the available literatures.[20,21]

Interestingly about the incidence of the rhinosporidiosis is that while several hundred people bathe in stagnant waters, only a few develop a progressive pattern of the disease. This might indicate the existence of predisposing factors in the host where the possibility of nonspecific immune reactivity in the host, blood group and HLA types has been suggested as important in the pathogenesis of Rhinosporidium seeberi in the establishment of an initial focus of infection.[20,21]

Since rhinosporidiosis has a slow course, lesions may be present for many years before the patients become symptomatic[7,20,21] and this appears similar to what was seen in our patient who reported a history of nasal obstruction and intermittent epistaxis for 2 years.

Rhinosporidiosis manifests as tumor-like masses, usually of the nasal mucosa or ocular, conjunctivae of humans and animals and patients with nasal involvement often have masses leading to nasal obstruction or bleeding due to polyp formation and it can spread to the nasopharynx, oropharynx, and the maxillary antrum.[1,3,20,21] The patient reported in our case had an isolated friable mass localized in the nasal cavity with no involvement of other anatomical sites such as maxillary sinus. The diagnosis is established by observing the characteristic appearance of the organism in tissue biopsies and computerized tomography (CT) scans. The lesion is friable, vascular pedunculated or sessile polyp, with a surface studded with tiny white dots due to spores beneath the epithelium, giving a ‘ strawberry-like ’ appearance. The lesion in our case report was friable evidenced by nasal bleeding upon probing.[20]

Systemic disease is rare but can include multiple mucocutaneous, hepatic, renal, pulmonary, splenic or bone lesions associated with fever, wasting, and even death.[20,21]

Though rare, spontaneous regression of Rhinosporidial growths has been noted in animals and in humans and therefore, medical and/or surgical intervention is necessary.[20,21] Wide local surgical excision of the Rhinosporidial growth is the treatment of choice to reduce the risk of recurrence, though this may be associated with significant morbidity due to hemorrhage and nasal septal perforation.[5,17] Therefore limited surgical excision and adjuvant medical therapies, including antifungals such as griseofluvin and amphotericin B, trimethoprim-sulphadiazine, and sodium stibogluconate have been tried with varied success. All drugs were endospore-static rather than endosporicidal. The strains obtained from human and animal rhinosporidiosis have shown genetic variations that might explain the variation of responses to some drugs though data on antimicrobial drug resistance in Rhinosporidium seeberi is lacking.[20,21] The only drug appearing to have clinical promise is Dapsone since it arrests the maturation of sporangia and promotes fibrosis in the stroma when used as an adjunct to surgery.[22] It could therefore be expected that pre-surgical Dapsone would minimize both hemorrhage by promotion of fibrosis as well as preventing the colonization and infection of new sites after the release of endospores from the surgically traumatized polyps.[23,24] Laser excision promises to be the mainstream treatment of sinonasal rhinosporidiosis in the future.[25] Our patient was kept on dapsone for 6-months after endoscopic nasal mass excision with no recurrence noted after 6-months of follow up. Our patient had complete nasal mass excision with wide surgical margins and cautery of the base of the Rhinosporidial growth endoscopically and was treated subsequently with Dapsone for 6-months.

## Conclusion

In Tanzania, rhinosporidiosis is non-endemic and in the Central zone, the disease is uncommon thus may pose diagnostic challenges. It’s therefore prudent for both clinicians and pathologists to have a high index of suspicion when managing patients with nasal masses even from non-endemic areas. Moreover, it will be very

crucial to make a follow up in the next few years particularly the clinical course of these patients to exclude the possibility of recurrence of the rhinosporidial growths after an extended period of time to evaluate its best treatment modality.

## ACKNOWLEDGEMENT

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## CONFLICTS OF INTEREST

None declared

## AUTHOR CONTRIBUTIONS

ZSA: Performed the surgery, collected information for the case and drafted the initial version of the manuscript. FZ performed histopathology and also provided critical feedback of the manuscript. AAK: Drafted the initial version of the manuscript. All authors read and approved the final version to be published in Clinical Case Reports

## ETHICS STATEMENT

This report is in accordance with the Declaration of Helsinki.

## CONSENT

The patients' mother gave a written informed consent prior inclusion of the child in this report

## FUNDING

None

## DATA AVAILABILITY

The data that support the findings of this report are available from the corresponding author upon reasonable request.

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