



Case Series

Rare Presentations of Cutaneous Rhinosporidiosis-A Case Series from A Tertiary Health Care Centre of Western Odisha

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ABSTRACT

Rhinosporidiosis is a non-neoplastic, chronic granulomatous inflammatory lesion caused by *Rhinosporidium seeberi*. More than 90% of cases are reported from Southeast Asian countries, with the highest prevalence in India and Sri Lanka. Although the nose is the common site, extra nasal manifestations are common in endemic regions. We present 9 cases of cutaneous rhinosporidiosis other than head & neck region, which were diagnosed preoperatively by cytology showing 100% concordance with histopathology.

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CASE SERIES-

Material & Method-

The retrospective study was conducted in the department of pathology, BBMCH, Bolangir, over a period of 7 years from May 2019 to April 2026. A total of 42 histopathologically confirmed cases of rhinosporidiosis were included. Out of 42 cases, 36 cases were preoperatively diagnosed by cytology followed by histopathological confirmation. Nine cases of skin nodules did not belong to head & neck region were diagnosed as cutaneous rhinosporidiosis by FNAC followed by histopathological confirmation. Two cases from the head & neck region and 3 cases where preoperative cytological evaluation was not done were excluded from the study.

FNA was done by using a 5 ml syringe with a 24gauge needle. The smears were prepared and stained with Diff Quik and Haematoxylin & Eosin. The cytological diagnosis was made by the demonstration of sporangia, including both intact and ruptured ones with endospores in an inflammatory background and cell debris. Then, corresponding biopsy samples were evaluated by routine histopathology. The H&E section showing characteristics feature of rhinosporidiosis such as double walled, refractile, acellular, and hyaline structure.

CASE PRESENTATIONS OF TWO CASES-

Case-1- A 43-year-old male presented with a firm swelling of size 11.0 x 6.0 cm over the distal part of the posterior left thigh. The patient was sent to the pathology department for cytological evaluation with a provisional diagnosis of soft tissue sarcoma. The FNA showed a cellular smear with the presence of multiple sporangia containing sporangiospores, ruptured sporangia with background showing sporangiospores, inflammatory cells, foreign body giant cells and cell debris.

The cytological diagnosis was cutaneous rhinosporidiosis. Then surgical excision was done, and the biopsy was received and processed in the histopathology section. The routine H&E section showed stratified squamous epithelium and the subepithelial tissues showing the presence of sporangia in different stages of maturation containing sporangiospores and a few atrophic and degenerated sporangia. The sporangia have a double-walled, refractile, acellular, hyaline structure. Good number of foreign body granulomas engulfing sporangia seen. The background showed mixed inflammatory cells.



Case-2- A 38-year-old male with a provisional diagnosis of fibroepithelial polyp in his left flank was subjected to cytological evaluation. The FNAC showed the classical features of rhinosporidiosis. The follow up biopsy showed cutaneous rhinosporidiosis with sporangia in different stages of maturation present within hyperkeratotic epidermis and subepithelial tissues.



RESULTS-

A total of 42 cases of rhinosporidiosis were encountered in the study in a period of 7 year. Out of these 42 cases, only 9 cases were diagnosed as cutaneous rhinosporidiosis by preoperative cytology, showing 100% concordance with follow up histopathology. The age range of these 9 cases were between 15 and 48 years. These 9 cases included 7 males and two females. The skin nodules of these 9 cases varying sizes from 1 cm to 11 cm. The sites of involvement were back (3 cases), posterior thigh (1 case), leg (2 cases), anterior abdomen (1 case), and left flank (1 case), and one disseminated case involving the right hand, back, and thigh.

Serial no.	Type of Haemoglobinopathy	No. of cases
1	Sickle cell Trait	143
2	Sickle cell Homozygous	117

3	Beta Thalassemia Trait	24
4	Compound heterozygous for Sickle cell – Beta Thalassemia	19
5	Beta Thalassemia major	07
6	Hereditary persistence of Fetal Haemoglobin(HPFH) Trait	02
7	HbJ Trait	02
8	Compound Heterozygous for HbS and HbE	01
9	Compound Heterozygous for HbS and HPFH	01
10	Compound Heterozygous for HbS and HbD Punjab	01
11	Compound Heterozygous for HbS and HbD Iran	01
12	Increase HbF as per age	05
14	Total no. of Haemoglobinopathy cases	323

Total No. of Cases 770			
Haemoglobinopathy Cases 323		Normal Cases 447	
Male 142	Female 181	Male 184	Female 263

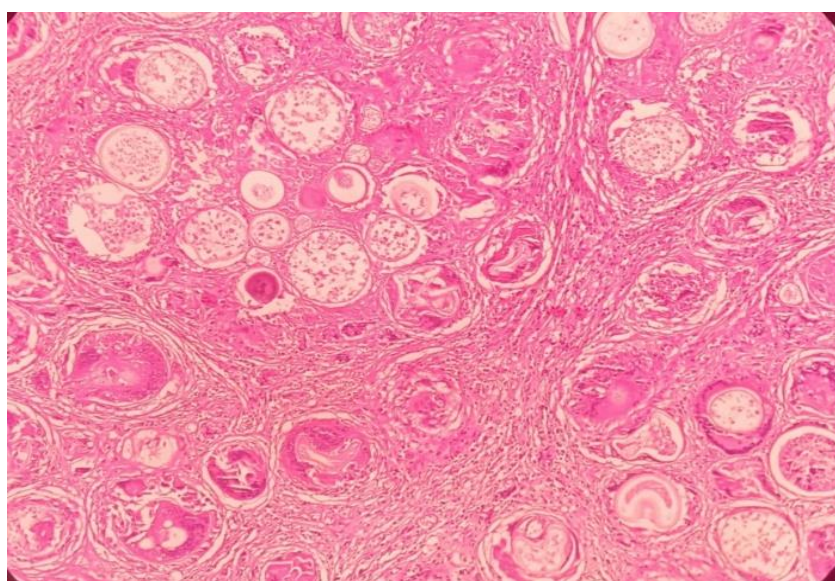


fig5 giant cell containing Sporangia, 400x, H&E

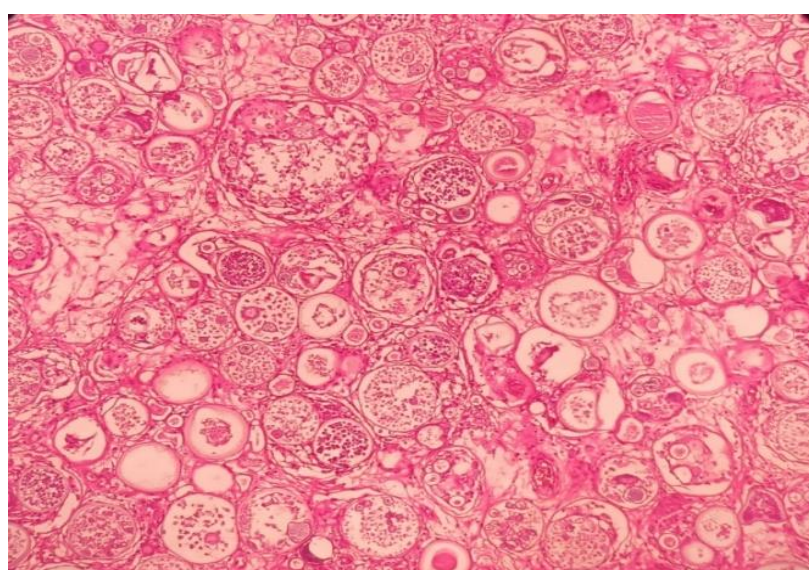


fig4 Sporangia in different stages of maturation,400X H&E Case1

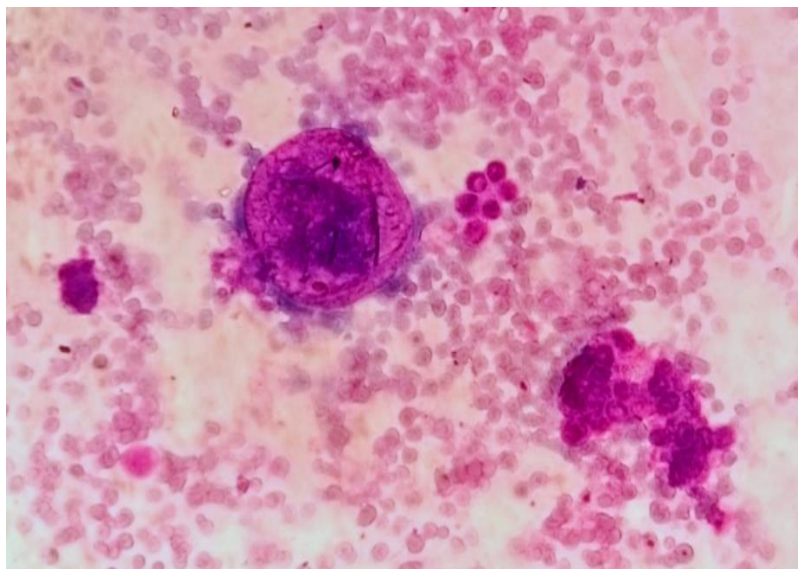


fig3, Cytology, Sporangia containing spores

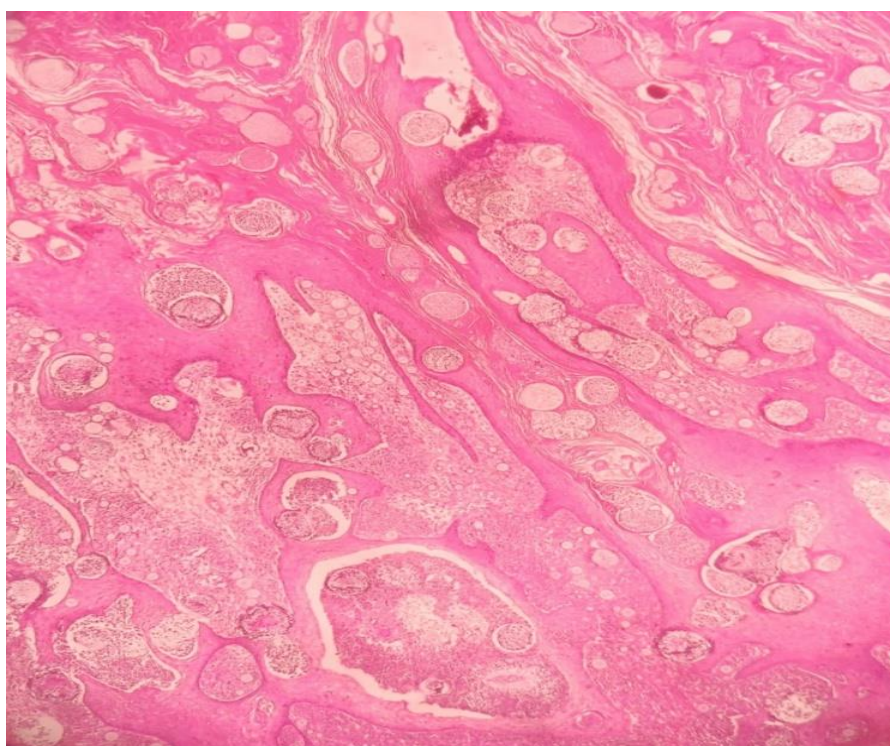


fig6 Sporangia in hyperkeratotic epithelium 200x, H&E

INTRODUCTION-

Rhinosporidiosis is a non-neoplastic, chronic granulomatous inflammatory lesion caused by *Rhinosporidium seeberi* (1). The 1st case was reported in the last part of the 19th century from Argentina (2,4). The term was coined by Ashworth in the year 1923(3). It was reported from more than 70 countries in the world, with maximum prevalence in Southeast Asia (1,4,5,6). Its taxonomy has changed over the years, and now it is considered a fungus-like aquatic protistan parasite belonging to the family Mesomycetozoa(1), which is not yet accepted universally (2,7). Nasal polyps are the most common presentation, followed by ocular swelling (8). Extranasal presentations of rhinosporidiosis are nasopharynx, larynx, trachea, palate, buccal mucosa, lip, skin, penis, urethra, vulva, and also bone. The organism enters the body through mucocutaneous junction or through injured epithelium. Till today, the organism is not isolated by any culture media, nor are there serological tests or imaging techniques available for the diagnosis (10). Routine histopathology is the gold standard for the definite diagnosis (11). Special stains like PAS, GMS, or mucicarmine only highlight the pathogen but are not essential for diagnosis (12). Preoperative diagnosis of rhinosporidiosis by FNAC or scrape cytology has been accepted as a rapid and accurate diagnostic procedure (13). Our study is to present the cases of cutaneous rhinosporidiosis other than the head & neck region diagnosed preoperatively by cytology, and confirmed by follow up histopathology.

DISCUSSION-

Rhinosporidiosis is caused by *Rhinosporidium seeberi* (1). The taxonomy of the organism is still uncertain. **Now** it is considered a fungus-like aquatic protistan parasite belonging to the family Mesomycetozoea (2,7). Mostly found in young adult males working in agriculture fields and fishery farming (1,4,9). The source of infection is from bathing in stagnant water bodies. Commonly presented as a polypoid, granular, friable mass in the nasal cavity (8), followed by an ocular presentation. Cutaneous manifestations are rare and varied, ranging from painless subcutaneous nodules to warty, ulcerative lesions on the skin. Large, subcutaneous swellings, sometimes even confused as soft tissue sarcoma clinically as in our first case. Histopathology is the gold standard diagnostic modality (11). The differential diagnoses are *Coccidioides immitis* and myospherulosis (12). The endospores of *Coccidioides* are smaller and fewer in number. Myospherulosis shows multiple Endo bodies in a parent body. Definite management is surgical excision (14) and in some cases Dapsone is prescribed with an aim to prevent recurrence (15).

CONCLUSION-

Cutaneous manifestations are not rare in this part of western Odisha. There is no characteristic presentation of cutaneous rhinosporidiosis, sometimes even confused with soft tissue neoplasm clinically. Preoperative cytological evaluation is a helpful tool for rapid and accurate diagnosis in guiding the proper management of cutaneous swellings. The study emphasizes the role of FNAC in subcutaneous swelling in high incidence region as in ours. The accurate preoperative FNAC assessment of rhinosporidiosis can achieve 100% concordance with histopathology.

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