

**International Journal of TROPICAL DISEASE
& Health**

24(1): 1-6, 2017; Article no.IJTDH.33978
ISSN: 2278-1005, NLM ID: 101632866

Cytodiagnosis of Disseminated Rhinosporidiosis - A Case Report

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Authors' contributions

This work was carried out in collaboration between all authors. Author RB reported the case and designed the study. Author MG did literature review, drafting and edits in the preparation of this report and is the corresponding author. Author VJ did literature searches and drafting of the manuscript. All authors read and approved the final manuscript.

Article Information

DOI: 10.9734/IJTDH/2017/33978

Editor(s):

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Complete Peer review History: <http://www.sciencedomain.org/review-history/19673>

Case Study

Received 7th May 2017
Accepted 15th June 2017
Published 23rd June 2017

ABSTRACT

Aim: Fine needle aspiration cytology of palpable lumps is a simple, inexpensive diagnostic modality. The aim of presenting this case is to highlight its utility in diagnosis of a patient presenting with multiple subcutaneous nodules.

Presentation of Case: We report a case of disseminated rhinosporidiosis diagnosed on fine needle aspiration cytology presenting with swelling in left supraclavicular region and similar swellings over his nose, oral cavity, chest wall, forearm and thigh.

Discussion: Rhinosporidiosis is a chronic granulomatous disease of the anterior nares and nasopharynx with polypoidal appearance. Subcutaneous tissue involvement with widespread nodules is extremely rare.

Conclusion: Disseminated Rhinosporidiosis is a rare presentation. Cytomorphological findings are distinctive; FNAC serves as a useful tool in early diagnosis and management of these cases.

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Keywords: *Rhinosporidium seeberi*; disseminated; recurrent; cytology; stains.

1. INTRODUCTION

Rhinosporidiosis is a chronic granulomatous disease caused by an organism of mesomycetozoa group known as *Rhinosporidium seeberi* [1].

The first case of rhinosporidiosis was described in 1900 by Guillermo Seeber presenting as nasal polyp and was considered as a fungus. Asworth described its life cycle in 1923 [2]. However, taxonomy of *R. seeberi* is still debatable and in last decade it was concluded that it is not a classic fungus, rather a human pathogen from Dermocystidium, a clade of aquatic protistan parasites [3].

Mucocutaneous sites are most frequently involved and present as polypoidal lesions in the nasal cavity. Orbital region, lip, palate, uvula, larynx, trachea, buccal cavity, lacrimal sac, scalp, skin, penis, urethra, vulva, and bone may also be involved. Disseminated rhinosporidiosis with involvement of visceral organs may be fatal. Infection of limbs is often associated with destruction of underlying bones [4,5].

Typical polypoid appearance of rhinosporidial lesions helps in correct preoperative diagnosis. However, atypical presentations with soft tissue masses may cause confusion. Aspiration cytology with demonstration of organism in its diverse stages with endospores and sporangia proves to be useful in such conditions. Similar morphological features are seen on histopathology [5,6].

Reported concordance of cytodiagnosis achieved 100% correlation with histology in a study by Sinha A et al. [5]. However, Pal S et al. [4] observed a sensitivity of 94% and specificity of 100%. One case was cytologically misinterpreted as suppurative inflammatory lesion.

FNAC is a reliable tool in preoperative diagnosis with few false negatives. Cytological diagnosis is specific and can be utilized as an initial diagnostic tool as in the present case of disseminated rhinosporidiosis presenting as subcutaneous tissue involvement with widespread nodules.

2. PRESENTATION OF CASE

A 32 years male presented with swelling in left supraclavicular region and right leg. He also complained of nose block and epistaxis. On examination, mobile, slightly tender subcutaneous nodules were also palpable over his jaw, chest wall and forearm. Overlying skin was normal at most places with a slight cutaneous protuberance over swelling in neck and on nose. Nasal and oral cavity examination showed fleshy hemorrhagic polypoid structure. (Fig. 1) Systemic examination was unremarkable.

Past history revealed that his illness started 14 years back as swelling protruding through the nose, for which he was operated. The swelling recurred at the same site after 3 months of operation. It was excised again and was diagnosed as rhinosporidiosis. He took treatment for the same and subsequent period was uneventful till he developed these swellings at multiple sites.

There was no past history of tuberculosis nor Diabetes Mellitus. No significant family history. There was no weight loss or loss of appetite.

Chest X-Ray was normal, HIV tests were negative, complete haemogram and urine routine were within normal limits. Serum immunoglobulin assay revealed increased IgG 27.0 gm/L (5-12) and Ig A- 5.42 gm/L (0.5-3.5), IgM was within normal limits 2.15 gm/L (0.3-2.3)



Fig. 1. Multiple subcutaneous swellings on (A) neck, (B) oral cavity and (C) lower limb

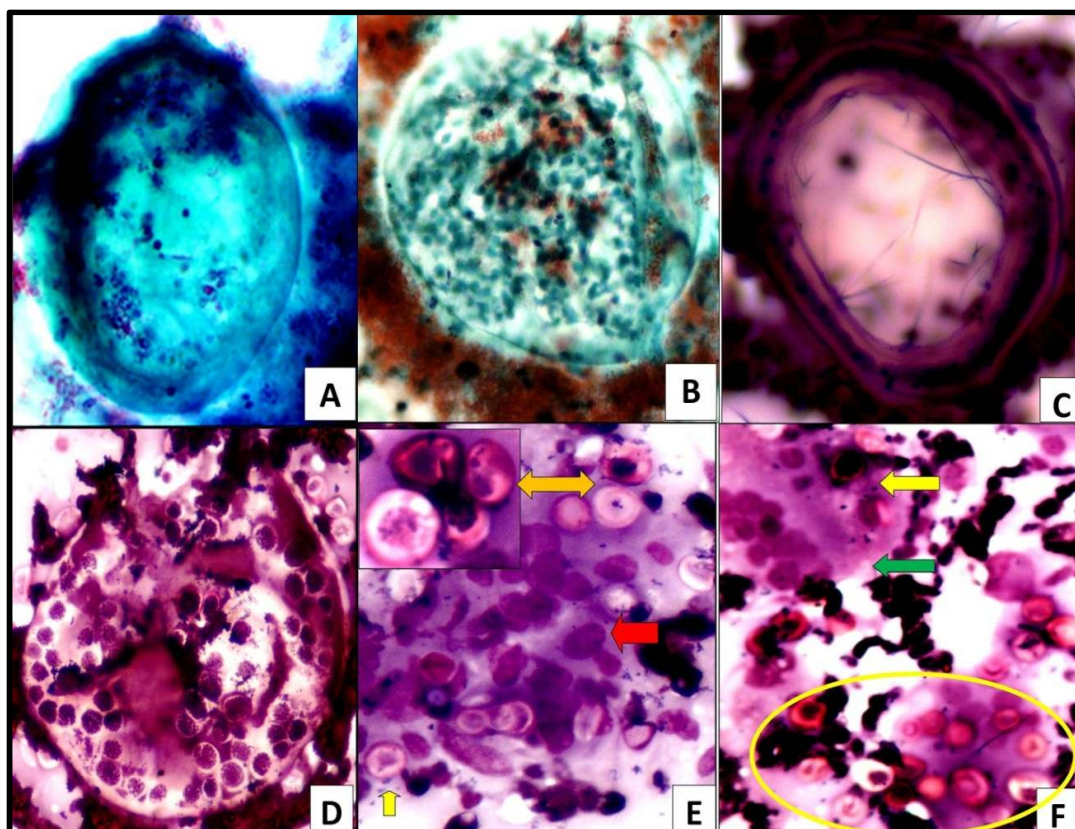


Fig. 2. Sporangia at various stages of maturation

A. Young trophic form (Pap, 400X) B. Globular sporangia containing endospores. (Pap, 400X) C. Sporangium with chitinous wall (MGG, 400 X). D. Ruptured sporangia releasing the endospores. (MGG, 400 X). E. Free endospores (yellow arrow and within inset (MGG, 1000X) with an epithelioid, granulomatous reaction (red arrow)(MGG, 400X). F. Giant cell (green arrow) engulfing endospores (yellow arrow) along with dispersed endospores (circle). (MGG, 100X)

3. METHODOLOGY

Fine needle aspiration cytology was done from different sites with 23-gauge needle with attached 10 ml syringe. Aspirate yielded thick blood mixed necrotic material. Both air dried and wet fixed smears were prepared. Direct examination of unstained smears revealed well circumscribed spherules with several brown endospores within. Air dried smears were stained with May-Grünwald-Giemsa (MGG) and wet fixed with papanicolaou stain. Few air dried smears were preserved for cytochemical stains due to presence of spherules on unstained smears. Cytochemical stains done were Periodic acid Schiff (PAS) and Toluidine blue. Aspirated material was also subjected to Concentration by 10% KOH solution and fungal culture. Morphometric analysis for assessment of diameter of sporangium and endospores was done on Motic image plus 2.0 ML software.

4. RESULTS

Microscopic examination of FNAC from all different sites revealed similar cytological material. There was presence of spherules in different stages of maturation (Fig. 2 A - D) and exhibiting great variability in size ranging from 50 to 300 μ m. Many ruptured sporangia and released numerous endospores ranging in size from 10.0 to 19 μ m (Fig. 2 E-F) were also seen. The spherule was refringent, the chitinous wall was unstained and the endospores were light blue or light green depending on whether stain was MGG or Pap stain respectively. Toluidine blue stained the spherules and endospore wall (Fig. 3A). PAS stain showed positivity for the chitinous wall and endospores with magenta staining (Fig. 3B).

Background showed dense acute inflammatory exudate with polymorphs arranged in rosette-like

pattern around most of the spores. Foreign body type of giant cells and granulomatous response were seen (Fig. 2 E-F).

Concentration by 10% KOH solution showed presence of multiple sporangia filled with endospores in various sizes. KOH destroys the epithelial cells but does not affect mycotic cells coated with chitin. However, fungal culture on Sabouraud dextrose agar of the aspirated material did not show any growth.

Based on the morphological features and findings of cytochemical stains, cytological diagnosis of disseminated rhinosporidiosis was made.

5. DISCUSSION

Disseminated rhinosporidiosis is rare [7]. However, review of published data reveals that the frequency of disseminated Rhinosporidiosis has increased in the last 20 years duration since first published case in 1955 [5,8,9].

Mode of spread may be either due to autoinoculation, hematogenous or lymphatic spread. They are of three types: Nasal polypoidal

lesions with satellite lesions, disseminated lesions with visceral involvement and primary cutaneous lesions without internal organ involvement [8,10].

Our case presented with a mixture of two types of lesions described by Hadke NS et al. [10]. He had nasal polypoidal mass with satellite lesions, muco-cutaneous lesions involving the oral cavity, and disseminated cutaneous nodules over neck, chest wall, forearm and right leg mimicking soft tissue tumour. Generalised involvement of skin is probably due to autoinfection caused by scratching the skin with fingers contaminated from infection site [11].

The first case of visceral involvement of Rhinosporidiosis was reported by Ranjam et al [12] in 1955 followed by Agarwal S et al. [11] in 1959 from India. Later there were few more reports of widespread cutaneous nodules together with nasal lesion. Nayak S et al. [9] reported one case of disseminated cutaneous rhinosporidiosis presenting with numerous subcutaneous swellings mimicking lipomas. Since then various authors have reported similar cases on histopathological examination. But number of cases reported on cytology is few in literature.

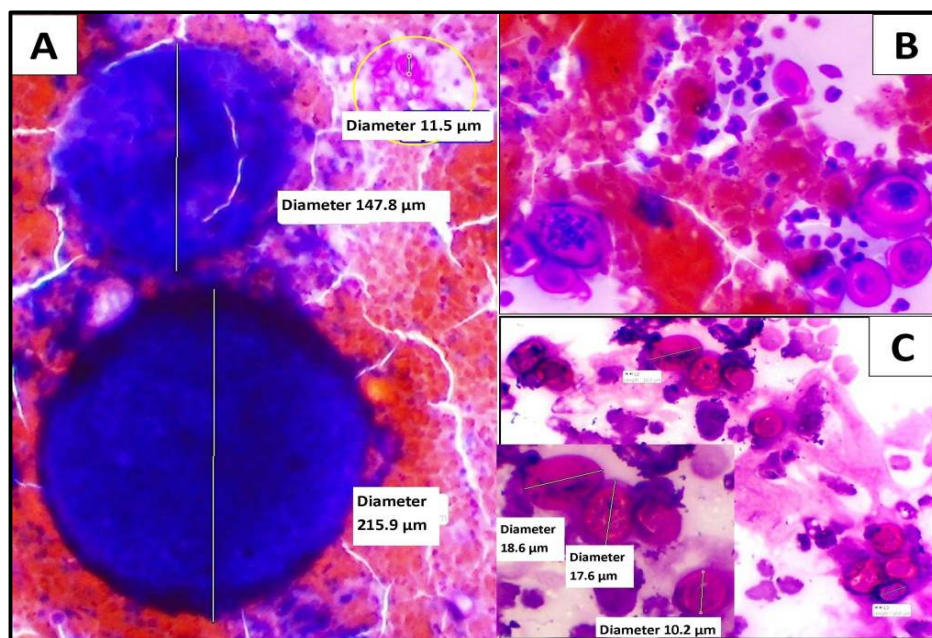


Fig. 3. Demonstration on cytochemical stains

A. Sporangium and dispersed endospores (yellow circle) (Toluidine blue stain 100, X with scale bar depicting diameter). B. Endospores with acute inflammatory infiltrate and hemorrhage (Toluidine blue stain 400, X) C. Dispersed endospores with mucoid material (PAS, 100X) (inset PAS, 400, X with scale bar) staining magenta, epithelial cells are negative for PAS

The cytologic features are typical. On direct examination the spherules are well-circumscribed, globular structures with several endospores within. The spherules show great variability in size, up to 10-fold. The diameter ranges from 30 to 300 microns. The presence of Electron Dense Bodies in the endospores is useful in confirmation of rhinosporidial identity. Commonly used stains for detecting *R. seeberi* include MGG and PAS stain [6,13,14]. Similar findings were observed in the present case. In addition we found that routinely done papanicolaou stain was also useful in demonstration of spherules and different stages of maturation. Also, toluidine blue helped in rapid diagnosis of organism. Documentation of definite findings on cytological examination may alleviate the need for histopathology especially in cases with dissemination.

The morphologic characteristics of *Rhinosporidium seeberi* resemble those of *Coccidioides immitis*. Both organisms have mature stages that consist of large, thick-walled, spherical structures containing smaller daughter cells (endospores) in a background of granulomatous reaction. Microscopically, rhinosporidium has sporangium of 50-1000 µm with endospores of 5-10 µm in the cytological smears and its inner sporangial wall stains with PAS. In contrast *Coccidioides immitis* are usually 20-80 µm in diameter, with thick, double refractile wall. The spherules are filled with small (2-4 µm in diameter) endospores [5].

Rhinosporidiosis remains a therapeutic challenge amenable only to surgical excision with a high risk of recurrence and occasional widespread and fatal complications. Surgical removal and electro desiccation are the treatments of choice. Dapsone may arrest the maturation of sporangia and accelerate degenerative changes in them [15].

6. CONCLUSION

Disseminated Rhinosporidiosis a rare presentation. Fine needle aspiration cytology serves as a useful tool in early diagnosis and may avoid surgical procedures especially in recurrent cases. Cytomorphological findings are distinctive and may be improved by use of various special stains.

CONSENT DISCLAIMER

As per international standard or university standard, patient's written consent has been collected and preserved by the author.

ETHICAL APPROVAL

It is not applicable.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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