

Case study

**CYTODIAGNOSIS OF DISSEMINATED
RHINOSPORIDIOSIS - A CASE REPORT**

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, which may have varied clinical differential diagnosis.

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.

Keywords : Rhinosporidiumseeberi, disseminated, recurrent, cytology, stains

INTRODUCTION

Rhinosporidiosis is a chronic granulomatous disease caused by an organism of the mesomycetozoea group known as Rhinosporidiumseeberi.[1]

23 The first case was described as nasal polyp by Guillermo Seeber from Buenos Aires
24 in 1900. The causative organism was considered as a fungus, and Asworth in 1923 described
25 its life cycle establishing the nomenclature *Rhinosporidiumseeberi*. [2]

26 It commonly involves the mucocutaneous sites presenting frequently as polypoidal
27 lesions in the nasal cavity. Uncommon sites of involvement include orbital region, lip, palate,
28 uvula, larynx, trachea, buccal cavity, lacrimal sac, scalp, skin, penis, urethra, vulva, and bone.
29 [3]

30 Rarely, disseminated infections are reported, involving limbs, trunks and viscera.
31 Brain involvement may lead to fatality. Disseminated infection of limbs is often associated
32 with destruction of underlying bones. [4]

33 Sporadic cases are reported from all over the world. But>90% of cases occur in South
34 East Asia especially from India, Sri Lanka and Pakistan. [1,3,4]

35 FNAC or scrape cytology is economical and reliable in preoperative diagnosis. FNAC
36 diagnosis of rhinosporidiosis is specific and has diagnostic utility in cases with unusual
37 clinical presentations. We report a case of disseminated rhinosporidiosispresenting as
38 multiple subcutaneous tissue involvement with widespread nodules diagnosed on fine needle
39 aspiration cytology.

40 **Presentation of Case**

41 A 32 years male presented with swelling in left supraclavicular region and right leg. He also
42 complained of nose block and epistaxis. On examination,mobile, slightly tender subcutaneous
43 nodules were also palpable over his jaw, chest wall and forearm. Overlying skin was normal
44 at most places with a slight cutaneous protuberance over swelling in neck and on nose. Nasal

45 and oral cavity examination showed fleshly hemorrhagic polypoid structure. (Figure 1)
46 Systemic examination was unremarkable.

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

52 There was no past history of tuberculosis, Diabetes Mellitus. No significant family
53 history. There was no weight loss or loss of appetite.

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

57 Fine needle aspiration cytology from different sites revealed similar cytological
58 material. Aspirate yielded thick blood mixed necrotic material. Both air dried and wet smears
59 were prepared. Direct examination of unstained smears revealed well circumscribed
60 spherules with several brown endospores within. Air dried smears were stained with Giemsa
61 stain and wet smears with papanicolaou stain. Microscopic examination revealed spherules in
62 different stages of maturation and exhibiting great variability in size ranging from 50 to 300
63 μm . Many ruptured sporangia and released spores were also seen. The spherule was
64 refringent, the chitinous wall was unstained and the endospores were light blue or light green
65 depending on stain. (Figure 2) Background showed dense acute inflammatory exudate with
66 polymorphs arranged in rosette-like pattern around most of the spores. Foreign body type of
67 giant cells and granulomatous response was seen.

68 Air dried smears were also stained with other cytochemical stains like Gridley,
69 Toluidine blue and Periodic acid Schiff (PAS). Gridley stain showed the chitinous wall
70 bright purple, and endospores brown. PAS stain showed positivity for the chitinous wall and
71 endospores outline. Toluidine blue also stained both the spherules and the endospore wall.

72 Concentration by 10% KOH solution showed presence of multiple sporangia filled with
73 endospores in various sizes. This destroys the epithelial cells but does not affect mycotic cells
74 coated with chitin. However, fungal culture of the aspirated material did not show any
75 growth. Based on all the morphological features and findings of cytochemical stains
76 cytological diagnosis of disseminated rhinosporidiosis was made.

77 **Discussion**

78 Disseminated rhinosporidiosis is rare.⁵ However, review of published data reveals that the
79 frequency of disseminated Rhinosporidiosis has increased in the last 20 years duration since
80 first published case in 1955.

81 Cutaneous lesions may occur due to autoinoculation or due to hematogenous spread
82 and are described to be of three types: Satellite lesions around the nasal polypoidal
83 lesions, disseminated lesions with visceral involvement and primary cutaneous lesions
84 without internal organ involvement.^[6,7] Our case showed a nasal polypoidal mass with
85 satellite lesions, muco-cutaneous lesions involving the oral cavity, and cutaneous nodules
86 over neck, chest wall forearm and right leg mimicking soft tissue tumour. Generalised
87 involvement of skin is probably due to autoinfection caused by scratching the skin with
88 fingers contaminated from infection site. Hematogenous spread produces visceral
89 involvement. ^[8]

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

96 The cytologic features are typical. On direct examination the spherules are well-
97 circumscribed, globular structures with several endospores within. The spherules show great
98 variability in size, up to 10-fold. The diameter ranges from 30 to 300 microns. Commonly
99 used stains for detecting *R. seeberi* include Giemsa, Gridley and PAS stain. [10,11] Similar
100 findings were observed in the present case. In addition we found that routinely done
101 papanicolaou stain was also useful in demonstration of spherules and different stages of
102 maturation. Documentation of definite findings on cytological examination may alleviate the
103 need for histopathology especially in cases with dissemination.

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

112 Rhinosporidiosis remains a therapeutic challenge amenable only to surgical excision
113 with a high risk of recurrence and occasional widespread and fatal complications. Surgical

114 removal and electrodesiccation are the treatments of choice. Dapsone may arrest the
115 maturation of sporangia and accelerate degenerative changes in them. [12]

116 CONCLUSION

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

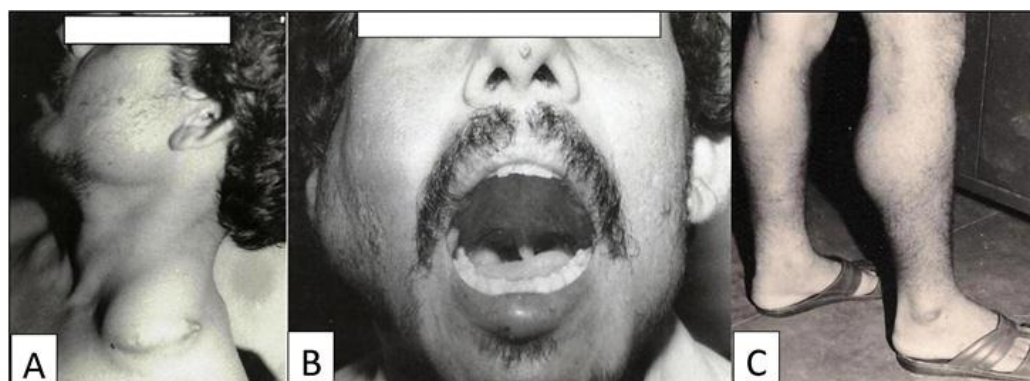
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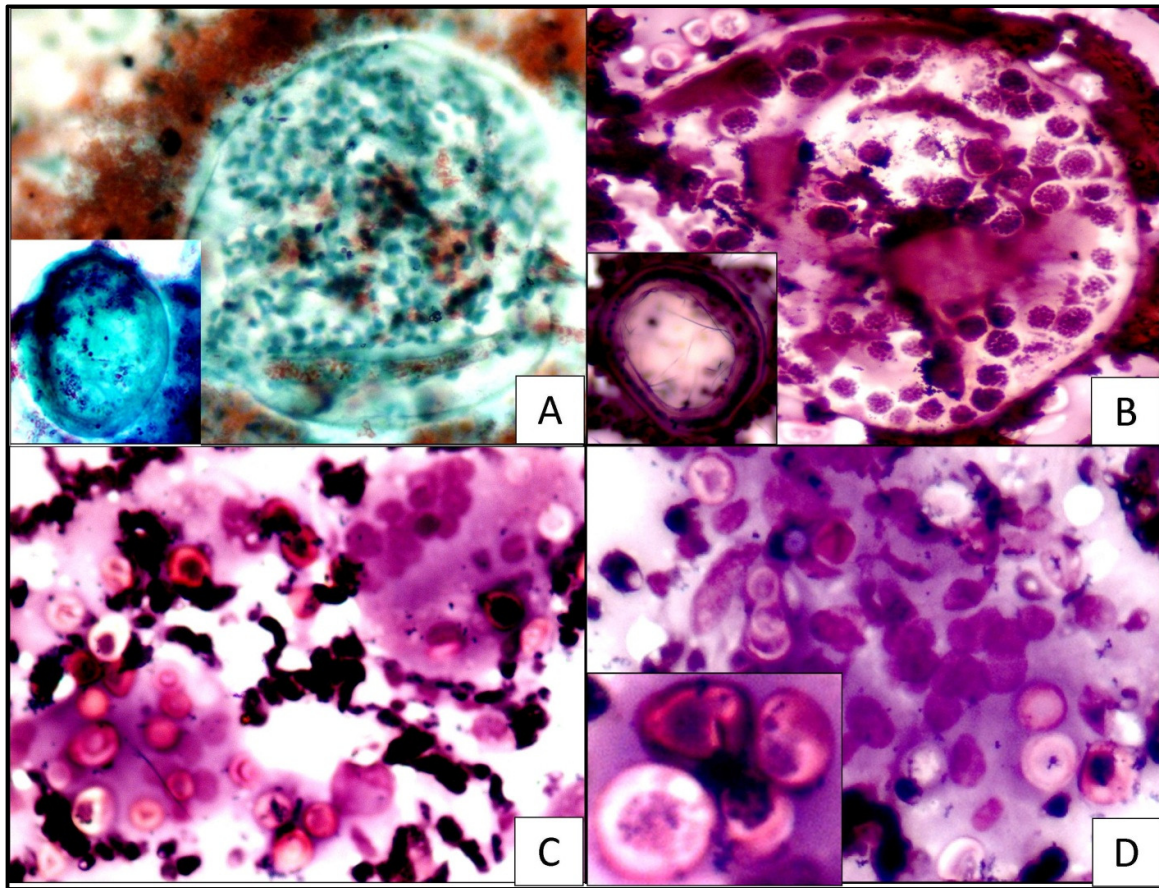
152 FIGURES

153 **Figure 1:** Multiple subcutaneous swellings on neck (A), oral cavity (B) and lower limb (C)



154
155 Figure 2: Sporangia at various stages of maturation. A. Globular sporangia containing
156 endospores. Inset - young trophic form.(Pap, 400X) B. Ruptured sporangia releasing the
157 endospores. Inset - sporangium with chitinous wall (MGG, 400 X). C. Giant cell engulfing

158 endospores. (MGG, 100X) D. Singly scattered endospores with an epithelioid, granulomatous
 159 reaction. (MGG, 400X) Inset –Endospores (MGG , 1000X)



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