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Case Report

Disseminated subcutaneous rhinosporidiosis masquerading soft tissue tumor: a rare presentation

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ABSTRACT

Rhinosporidiosis is a chronic granulomatous disease caused by *Rhinosporidium seeberi*. It primarily affects nasal cavity. Cutaneous manifestation of the disease is rare. Here we report a case of disseminated subcutaneous rhinosporidiosis, clinically and radiologically suspected to be a soft tissue tumor, which was diagnosed by FNAC and confirmed by histopathological examination.

Key words: Rhinosporidiosis, Disseminated, Subcutaneous

INTRODUCTION

Rhinosporidiosis is caused by a pathogen *Rhinosporidium seeberi*, is highly endemic in South Asia especially in south India and Srilanka, and was first described in Argentina by Seeber in 1900. It commonly affects the upper respiratory sites anterior nares, nasal cavity, nasopharynx, larynx, and soft palate, Ocular sites (15%).[1,2] Cutaneous and subcutaneous lesions are infrequent and are generally associated with mucosal lesions.[3] Here we report a case of Disseminated Rhinosporidiosis that presented with multiple subcutaneous nodular masses, which underwent FNAC and was confirmed with histopathology.

CASE REPORT

A 52 year old male, presented to the general surgery outpatient department with multiple swellings over the right forearm, right thigh, right leg and left leg for the past one year. The swellings were soft to firm in consistency, largest measuring 7cms in the right thigh (Figure 1) and the smallest measuring 2 cms in the right forearm. There was no regional lymphadenopathy. Other investigations included complete hemogram, liver function test, renal function test, plasma glucose were normal, HIV serology by ELISA was negative. USG and MRI of the mass were done and suggested possibility of neurofibromatosis with cystic component in largest mass. Mass was fairly circumscribed in subcutaneous plane with no invasion in to the underlying muscle or bone. A provisional diagnosis of neurofibromatosis / lipomatosis was made clinically and he was referred for FNAC.

Figure 1: Nodular masses in the right thigh and leg and left leg.

Aspirate was taken from different masses, thigh mass revealed 2ml of straw colored fluid which was subjected to sediment cytology. Smears revealed globular sporangia with spores, chronic inflammatory cells, and few foreign body giant cells (Figure 2).

So a diagnosis of disseminated subcutaneous rhinosporidiosis was made. Subsequent evaluation to rule out visceral organ involvement was done with ultrasound abdomen, CT-Brain, X-Ray chest. No visceral lesions were found. ENT examination revealed an Oro-pharyngeal mucosal lesion which was clinically mistaken for malignancy.

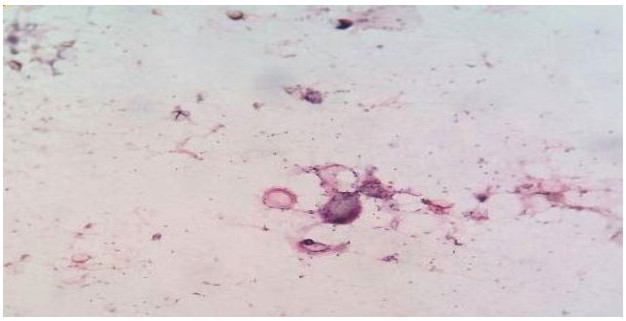

Figure 2: FNAC smear showing sporangia in different stages of maturation with inflammatory infiltrate. H&E stain, 100X.

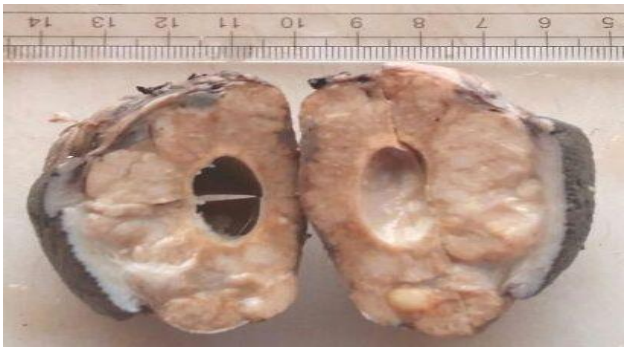

Figure 3: Excised mass showing subcutaneous yellowish white nodule with cystic change.

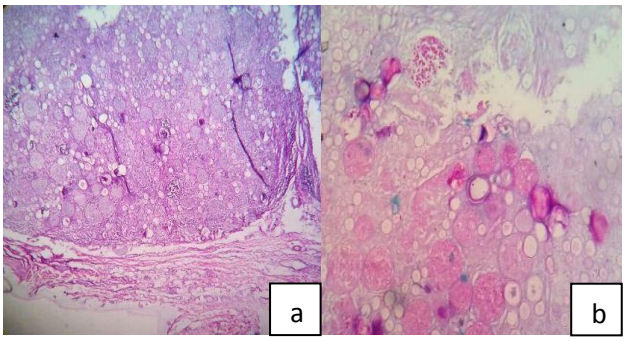

Figure 4: Photomicrograph of rhinosporidiosis showing sporangia and spores; (a) H&E stain, 40X; (b) PAS stain, 40X.

All lesions were excised; grossly they were fairly circumscribed subcutaneous masses, creamy yellow and soft in consistency with cystic change in largest mass (Figure 3) and subjected to histopathological examination for confirmation. Microscopically, numerous sporangia in different stages of maturation with endospores which stained magenta with PAS were seen in the subcutaneous plane (Figure 4a and b).

DISCUSSION

Rhinosporidiosis presents as a granulomatous inflammation of the mucocutaneous regions of the nose, conjunctiva, oro-naso-pharynx, skin, genito-urinary sites as well as in subcutaneous areas. Tropical ecological nature of the soil and water has been implicated in its endemicity. The natural habitat being ground water in lakes, ponds, and river beds containing sand; exposure to soil and soil dust is the main cause seen in them.[4]

The mode of infection of *R. seeberi* is usually by way of traumatized epithelium called transepithelial infection, most commonly in nasal sites. Other modes are auto-inoculation especially after surgical removal of polyps which cause spillage of spores in the adjacent epithelium. Hematogenous mode of spread has been implicated for distant and disseminated spread to various other sites like limbs and extremities with the development of subcutaneous granulomata without any breach of the overlying skin from subclinical foci of infection in the upper respiratory area, as seen in our case.[1,3]

Cutaneous lesions are rare and if present are generally associated with mucosal lesions. Three main types of cutaneous rhinosporidiosis have been implicated:

- Satellite lesions associated with nasal involvement;
- Disseminated skin lesions with or without nasal involvement; and
- Primary cutaneous type.[5]

Cutaneous lesions present as masses mimicking warts, neurofibromas, lipomas, verrucous tuberculosis, pyogenic granuloma or even malignant masses.[5-7]

Subcutaneous lesions are not vascular and could be excised with minimal bleeding and the masses have cerebriform cream colored appearance, with pale yellow cut surface and the consistency of brain tissue. Cystic change can occur and the fluid contains sporangia.[6] Our case had similar gross findings with cystic change in the largest mass (Figure 3).

FNAC of the Cutaneous masses reveals intact spherical sporangia filled with endospores and dispersed spores with few foreign body giant cells and chronic inflammatory infiltrate.[2,6] Cytological smear shows well circumscribed spherules seen as globular structures with endospores within it. The spherules diameter size ranges from 30 to 300 microns. The spores stained magenta with

PAS, a feature used to differentiate them from epithelial cells (PAS negative).[2] We had similar findings in our cytology smears. The definitive diagnosis is by histopathological examination showing the organism in its diverse stages as well as demonstration of the large thick walled sporangia, seen as spherical structures containing smaller sporangiospores, along with chronic inflammatory cells, macrophages, neutrophils, and with numerous extruded endospores lying freely in a fibrous or fibro-myxoid background. Mature sporangia have an operculum extruding the spores out.[2]

The various developmental stages of the sporangia can be readily identified by routine hematoxylin and eosin stain.[2,5] Rhinosporidiosis must be differentiated from *Coccidioides immitis*, the latter having smaller spherules 20-80 microns in size, the endospores are 2-4 microns in diameter.[2] Surgical excision is the mainstay in treatment for masses. Recurrence, dissemination in anatomical close sites and secondary bacterial infections are the most frequent complications.[7]

CONCLUSION

To conclude, cutaneous rhinosporidiosis should be borne in mind, when dealing with cases presenting as multiple cutaneous nodules over extremities in endemic areas. Fine needle aspiration is distinctive in these unusual presentations and a definitive diagnosis can be made easily and rapidly in such cases.

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