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DISSEMINATED CUTANEOUS RHINOSPORIDIOSIS - A CASE REPORT

DIVYA
Department of Microbiology,MADURAI MEDICAL COLLEGE AND HOSPITAL

Abstract : Rhinosporidiosis is a chronic granulomatous infection caused by Rhinosporidium seeberi, characterized by polyposis of nasal cavity, conjunctiva and other body sites. Rhinosporidiosis has been reported from many countries but is endemic in certain parts of India and Sri Lanka. It is also known to involve many rare sites and may become disseminated to occur in generalized form. The skin manifestations in rhinosporidiosis are rare. This is a case report of a patient who presented with disseminated cutaneous rhinosporidiosis and R.seeberi was diagnosed with Potassium hydroxide mount, culture in sabouraud's dextrose agar and by Histopathological examination. Patient has been treated with surgery, Diathermy and LASER therapy for his nasal polyps and cryotherapy for his cutaneous lesions. Dapsone has been given as systemic therapy.

Keyword : Cutaneous, Disseminated, Histopathology, Potassium Hydroxide mount, Rhinosporidiosis.

DISSEMINATED CUTANEOUS RHINOSPORIDIOSIS - A CASE REPORT ABSTRACT:

Rhinosporidiosis is a chronic granulomatous infection caused by Rhinosporidium seeberi, characterized by polyposis of nasal cavity, conjunctiva and other body sites. Rhinosporidiosis has been reported from many countries but is endemic in certain parts of India and Sri Lanka. It is also known to involve many rare sites and may become disseminated to occur in generalized form. The skin manifestations in rhinosporidiosis are rare. This is a case report of a patient who presented with disseminated cutaneous rhinosporidiosis and R.seeberi was diagnosed with Potassium hydroxide mount, culture in sabouraud's dextrose agar and by Histopathological examination. Patient has been treated with surgery, Diathermy and LASER therapy for his nasal polyps and cryotherapy for his cutaneous lesions. Dapsone has been given as systemic therapy.

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INTRODUCTION

Rhinosporidiosis in humans has been known since 1892 and has been reported from over 70 countries including India and Sri Lanka where the disease is hyperendemic. In India, the disease is endemic in the states of Chattisgarh, Kerala, Tamil Nadu, Orissa and West Bengal. Nearly 90% of cases of rhinosporidiosis are from India and Sri Lanka[(1)].

Rhiniosporidiosis is a chronic granulomatous infection involving the mucous membranes. The most common site of involvement is the nose, nasopharynx where the lesion presents as a polypoidal mass. The next common site is the eye involving the conjunctiva. Lesions of rare sites have been reported such as bone, vagina, oropharynx, buccal mucosa, tongue, paranasal sinuses, skin and subcutaneous tissue, penis, urethra, trachea, larynx and bronchus. Rarely disseminated and generalized rhinosporidiosis occurs [(12)]. The primary lesion in most of the patients is in the nose.

This is a case report of a patient who had nasal polyp for the past thirty years and has presented with mass in the oral cavity and papules, nodules, plaques, hyperkeratotic cutaneous horns and furunculoid lesions over face, trunk, back and lower limbs for the past 1 year. Patient gives history of surgery done for his nasal polyp and gives history of taking bath in community pond. This case is presented for its rarity in our locality. Diagnosis is done by Potassium Hydroxide mount and confirmed by Histopathology.

CASE REPORT:

A 48 year old male from Rajapalayam, Labourer by occupation, presented to Govt. Rajaji Hospital, Madurai with papules, nodules and hyperkeratotic skin lesions over back, trunk, face and lower limbs for the past 1 year. History of mass in the oral cavity is present for the past 1 year which developed gradually to the present size. Patient gives history of nasal polyp for the past 30 years. No history of pain or bleeding from the nasal or oral cavity lesions**. Patient gives History of taking bath in community pond frequently.**

He has been diagnosed to have nasopharyngeal rhinosporidiosis by potassium hydroxide mount which revealed thick walled sporangia with endospores and confirmed by histopathology. CT scan contrast studies also showed the mass enhanced with contrast, showing polypoidal soft tissue mass in the inferior turbinate prolapsing into nasopharynx and blockade of osteomeatal units confirming nasopharyngeal rhiniosporidiosis.

Patient has been treated surgically for his condition. Few months later again he had similar polypoidal mass in his nose. In the past 10 years he had recurrences of nasal polyps and had been operated for 7 times for his nasal polyp, followed by which he developed variety of skin manifesitations in the past 1 year.

LASER therapy was given for his nasal polyps at last and his nasal polyps regressed.

On local examination, multiple soft to firm skin coloured nodules of varying sizes on lateral aspect of right eye, root of nose, left inner thigh and dorsal aspect of right hand were present. Multiple warty papules and cutaneous horn like lesions, crusted plaques, ecthymatous lesions and healed scars were seen over fore arm,back and lower limbs. (Figure.2a,2b,2c,2d,2e). Examination of oral cavity showed circumscribed reddish granulomatous growth of 4×3 cm size, studded with white spots over the hard palate (Figure.1). Examination of other mucosae is normal. General and Systemic examination revealed no abnormalities. A provisional diagnosis of recurrent nasopharyngeal rhinosporidiosis with cutaneous dissemination has been made.

Potassium Hydroxide mount of scraping from the surface of nodule over the palate and from aspirates of cutaneous lesions (nodules and furunculoid lesions) revealed multiple thick walled sporangia with numerous endospores (Figure.3a & 3b). Biopsy taken from oral mass and nodular lesions of skin also confirmed rhinoporidiosis histopathologically (Figure.4a&4b). **Culture** in sabouraud's dextrose agar **showed no growth** even after 6 months. **Serology for HIV is non-reactive**.

The patient has been treated with cryotherapy for his skin lesions and diathermy and LASER therapy for his oral cavity mass and nasal polyps. Dapsone has been administered as systemic therapy.

DISCUSSION:

Rhinosporidiosis is a chronic, usually painless, localised granulomatous disease of mucous membranes affecting humans and animals[4] and was first described in Argentina[1].**The disease is endemic in certain parts of the world. In Tamil Nadu, the disease is endemic in southern districts such as Madurai, Rajapalayam, Ramnad and sivaganga[9]. Our patient is from Rajapalayam having a strong predisposition to acquire the disease. The reason for the endemicity may be attributed to the physical and chemical properties of the stagnant water and the synergistic action of some aquatic organisms in the water which is the source of infection to humans. Some of the host factors that predispose are blood group O (70%) followed by AB, as reported in few literature [9].** The causative organism Rhinosporidium seeberi has been considered to be a fungus for long time and has never been cultured. Based on sequence analysis of 18S small subunit ribosomal DNA of the organism, it has been reclassified and included among mesomycetozoa, an aquatic protistan parasite (clade of fish parasites) that form a branch of the evolutionary tree near the animal–fungus divergence [1], **the DRIPs clade** (*Dermocystidium Rosette Agent Icthyophonus Porospermium)* and hence called as pseudofungal organism. Herr et al,[2] recently proposed that the organism should be considered in a new eukaryotic group of protists known as Mesomycetozoa.

Primary lesion occurs in the nose and nasopharynx more commonly, the ocular structures such as conjunctiva less commonly, and the skin and subcutaneous tissue more rarely. **Cutaneous lesions are infrequent and are generally associated with mucosal lesions as in our case, and also as described by Rivard et al.,[10]. Oral cavity mass was present in our patient similar to the case report of shenoy et al**[11]. Three types of skin lesions can occur (1) satellite lesions, in which skin adjacent to the nasal rhinosporidiosis is involved secondarily; (2) generalized cutaneous type with or without nasal involvement, occurring through hematogenous dissemination of the organism; and (3) primary cutaneous type associated with direct inoculation of organisms on to the skin [2]. Apart from these a possibility of lymphatic spread has also been considered[1]. Disseminated disease is quite rare [3].

A case of disseminated cutaneous rhinosporidiosis in **HIV seropositive patient** has also been described by **Sahoo and colleagues**[8]. **Stricker** and colleagues described in a patient of **AIDS** in literature. **Tolat et al.,[15], and Kumari et al., [3]** described **disseminated cutaneous rhinosporidiosis with** systemic pulmonary involvement with a cutaneous horn secondary to rhinosporidiosis in an **immunocompetent male. Our patient also tested negative for HIV serology** who had cutaneous dissemination but **without** any systemic involvement.

There may be wart like, papillomatous or sessile masses in areas adjoining to nose and face.

There may be subcutaneous scattered nodules [13], which later on ulcerate and are seen fungating over skin. The cutaneous lesions are rarely pedunculated, the mode of infection may be direct inoculation to the local site or it may be through hematogenous dissemination resulting in painless, firm to hard, subcutaneous nodules that remain unattached to skin. The subcutaneous rhinosporidiosis resembling a tumour is described by various adjectives as "giant cutaneous", "tumoral disseminated" and "generalized one" or "rhinosporidioma" [14].**Our patient had varying cutaneous manifestations such as papules, nodules, furunculoid, ecthymatous and verrucous lesions which coincided with case report of Kumari et al., except for the satellite lesions near the nasal polyps which were absent in our case suggesting a role for hematogenous dissemination rather than a contiguous spread from nasal lesion.** Vijaikumar M et al., **[3] described a verrucous lesion of palm, whereas our patient had the cutaneous horn like lesion on the trunk.**

The habitat of the organism is fresh and stagnant water coming frequently in contact of man & animals. R. Seeberi is believed to be a hydrophilic organism. It is a very common practice among people residing in endemic areas to have a **dip in holy ponds** especially during festival seasons. This could be a major risk factor for transmission of disease possibly from one person to other **as suggestive strongly in our patient**. **Kumari** et al also reported a case with similar history of the patient having a dip in ponds and had disseminated cutaneous rhinosporidiosis and **also described the possibility of transmission to humans by direct contact with spores through dust, through infected clothing or fingers [3].**

. The diagnosis of Rhinosporidiosis can be made by KOH mount as done by **shenoy et al**.,[11] and confirmed by histopathological examination by using H & E stains. Special stains such as Gomori Methanamine Silver, PAS and Mucicarmine can be used for better visualization of the organism. Histopathological examination shows the epithelium is hyperplastic with papillamatosis and pseudocysts may be formed. Numerous globular cysts of varying shape, representing sporangia in different stages of development with mixed inflammatory infiltrate with lymphocytes and histiocytes are seen[5]. It is difficult to determine the drug sensitivity, since growing the organisms in vitro or in vivo has been a failure [1]. The cultivation of *R. seeberi* was performed by Levy *et al,* [6] but was not confirmed by other researchers [7].Rhinosporidium seeberi can be differentiated from other organisms like coccidioides immitis by the morphology and special stains. R.seeberi forms round, thick-walled cysts (sporangium) in the submucosa, varying in diameters from 10-100 µm, (whereas the spherules of coccidiodes measure about 30-60 µm) often visible through the mucosa as white dots [12]. Mature cysts become filled with numerous endospores, which on release become new cysts.

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Spontaneous regression of lesions can rarely occur. However, it should be treated early in order to prevent extension of lesions or dissemination. Diathermy excision with cauterization of base is the treatment of choice, inspite of which recurrences have occurred. Systemic therapy with Dapsone alone is proven to be somewhat effective. This patient has been treated with diathermy excision and LASER therapy for his nasopharyngeal lesions and cryotherapy for his cutaneous lesions. Dapsone 100 mg once daily has also been advised as systemic therapy for one year and the patient was followed up regularly. Inspite of surgical & systemic therapy patient had recurrences. Development of cutaneous lesion may be an early sign of dissemination. **There are only so far two case reports with cutaneous horn like lesions, hence this case is reported for its rare presentation in our locality. This case report highlights the rare occurrence of varied manifestation of cutaneous rhinosporidiosis, the possibility of which should be kept in mind when a patient with polypoidal mass in the nasal cavity presents with cutaneous lesions**

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Fig .1 Mass in the oral cavity

Fig 2a-Nodular lesion in left thigh

Fig .2b warty papules and cutaneous horn like lesions over trunk,abdomen and fore arm

Fig.2a- Nodular lesion near Right Eye

Fig .2c crusted plaque over left leg

Fig .2d Ecthymatous lesion over back

Fig .2d ecthymatous lesion over left elbow

Fig .2e Healed scar over left fore arm

Fig .3a-KOH Mount-Sporangium with endospores

Fig .3b KOH mount.

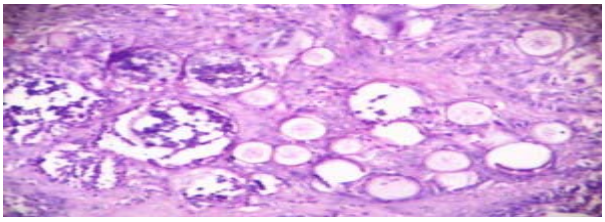

Fig .4a Histopathology showing sporangium with endospores

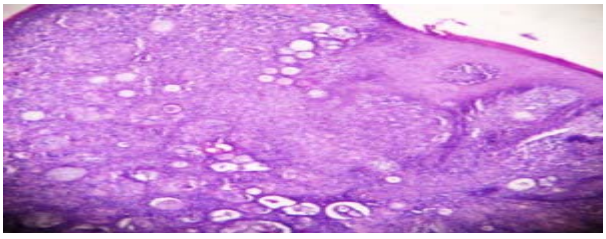

Fig .4b Histopathology showing epidermal hyperkerato-sis,sporangium with endospores