



Disseminated rhinosporidiosis- a rare case presentation

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ABSTRACT

Rhinosporidiosis is a granulomatous disorder caused by *Rhinosporidium seeberi*. It usually occurs in the mucous membranes of nose, nasopharynx, eyes and less commonly in extra nasal sites such as skin, bones, genitalia and even the internal organs. We hereby report a case of 25 year old immuno competent male who presented with nasal obstruction along with a subcutaneous nodule over ulnar aspect of right forearm and infected wound over lateral malleolar area of right leg. On examination, a pinkish polypoidal mass was found inside left nostril. Incisional as well as excision biopsy confirmed the lesions as disseminated rhinosporidiosis. The patient was treated with excision and dapsone therapy. After 5 months of therapy, the patient has disease-free survival. We report this rare case of rhinosporidiosis with disseminated subcutaneous as well as involvement of bone.

INTRODUCTION

Rhinosporidiosis is a chronic granulomatous infection caused by *Rhinosporidium seeberi*. It is endemic in India and Sri Lanka, also has been reported from United States, South America and Iran. [1-3] Infection is usually caused by contact with fresh water such as swimming pools. Rhinosporidiosis frequently involves the nasopharynx (70%) presenting as a painless, friable, polypoidal growth which may hang anteriorly into the nares and posteriorly into the pharynx. The conjunctiva and the lacrimal sac are involved in 15% of cases. Occasionally, rhinosporidiosis affects the lips, palate, uvula, maxillary antrum, epiglottis, larynx, trachea, bronchus, ear, scalp, vulva, vagina, penis, rectum, and the skin.[4] Involvement of bone is rare.

CASE REPORT

In May 2013, a 25 year old man presented with a 5-month history of an ulcerating and fungating mass over the right lateral malleolar area, subcutaneous nodule over ulnar aspect of right forearm for last 8 months along with left nasal obstruction for last one and half year. There was sero-sanguinous foul smelling discharge from the swelling over lateral malleolus, which bled on touch. The mass was 9x7x5 cm in dimension, firm, friable, and multi-lobulated with a delineated margin. There was no pain over the swelling and the patient had no fever. Nasal cavity examination shows pinkish polypoidal mass in left nostril and bled on touch. His complete hemogram, blood sugar, liver and renal functions tests were within normal limits. Serology for HIV, HBsAg and HCV were negative.

Radiography of right ankle joint revealed an expansile, lytic, destructive lesion involving the calcaneum. Fine needle aspiration from the swelling over right forearm yielded 20ml. of blood mixed fluid. Fine needle aspiration from right malleolar area produced bloody aspirate.

Cytological analysis showed plenty of polymorphs, lymphocytes, histiocytes, few foreign-body giant cells and sporangia with endospores. Few endospores were also present dispersely in the background. The background was dirty with presence of RBCs. Excisional biopsy of the nasal mass, forearm mass as well as from calcaneum confirmed the diagnosis to be rhinosporidiosis.

DISCUSSION

Rhinosporidiosis is known for over a hundred years and was first described in Argentina. The etiologic agent, *Rhinosporidium seeberi*, is closely related to several fish pathogens. It is an aquatic protozoan and was previously considered to be a fungus; however its taxonomic position is unclear. The infection commonly affects nasal mucosal membranes and ocular conjunctiva of humans & animals, producing slowly growing masses that degenerate into pink to purple & friable polyps. Four forms of the disease are recognized i.e. nasal, ocular, cutaneous & disseminated (rare). Infection of nose & nasopharynx is observed in 70% of patients of rhinosporidiosis; infection of palpebral conjunctiva or associated structures (lacrimal apparatus) is observed in 15%. Cutaneous lesions are infrequent & are generally associated with mucosal lesions. Involvement of bone is rare. Isolated osteolytic lesions have been reported involving the talus,[5] tibia,[6],[7] femoral



Fig. 1. Figure showing rhinospore mass in left nostril.



Fig. 2. Figure showing subcutaneous nodule in right forearm and infected wound over lateral malleolus.



Fig. 3. X-ray showing osteolytic lesion involving calcaneum.

condyle,[8],[9] hand and feet, [10] calcaneum, [11] and clavicle.[12] Dissemination to anatomically unrelated sites is mainly attributed to hematogenous spread.[13] Direct implantation by contaminated nasal secretions either during trauma or surgery can also implant the spores and cause secondary lesions.[13]

Rhinosporidiosis has been reported from several countries but is most commonly reported from India and Sri Lanka. Apart from human, this aquatic micro-organism can also cause disease among the farm, domestic & wild animals. Inoculation of injured epithelium from the natural habitat seems to be the mode of transmission. Some individuals are more prone to develop this infection than others due to some unknown host factors.

Several modes of spread have been postulated for cutaneous rhinosporidiosis. Surrounding cutaneous surfaces may be involved due to a contiguous spread from nasal mucosa. Hematogenous dissemination can lead to generalized cutaneous lesions. Direct inoculation of the organism results in primary cutaneous type. Apart from this, a possibility of lymphatic spread has also been considered.

Spontaneous regression of lesions can rarely occur, however this should be treated early in order to prevent extension of lesions or dissemination. It is difficult to determine the drug sensitivity since growing the organisms in vitro or in vivo has been a failure. Diathermy excision is the treatment of choice, but in spite of this, recurrences have occurred. Several drugs have been tried but dapsons is the most frequently used one. But, the efficacy is ambiguous. Its probable mode of action is to arrest the maturation of the sporangia and to induce stromal fibrosis.

CONCLUSION

In conclusion, this is an infrequent human case of rhinosporidiosis. Rhinosporidiosis is a condition which both clinicians and pathologists should keep in mind while dealing with patients from endemic countries with nasal masses. Moreover, it will be very interesting to follow the clinical course for our patients. An eventual recurrence of the lesion, usually occurring after a long time, would mean a true relapse, excluding the possibility of a re-infection, more probable in the endemic areas.

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