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Case Series

Cutaneous Rhinosporidiosis: A Rare Case Series

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Abstract

Background: Rhinosporidiosis is a chronic granulomatous infection caused by Rhinosporidium seeberi. Most commonly it affects male population and involves the mucous membranes of the nostrils, nasopharynx and eyes. Involvement of cutaneous locations including lid and cheek has been seldom given account of. Here we are presenting a case series of ten cases of Rhinosporidiosis over cutaneous sites. The disease responds well to wide local surgical excision and follow up treatment with Dapsone.

Material and Method: In this case series we have included all the cases of cutaneous rhinosporidiosis which were observed in the past three years in tertiary medical health care centres of Chhattisgarh.

Results: Although rare, it is not so uncommon to find Rhinosporidium seeberi on cutaneous sites. Chhattisgarh is primarily a tribal area and due to lack of sanitation and clean water facilities cutaneous rhinosporidiosis was diagnosed in ten patients in this study. In our study the age ranged from 2nd -6th decade. It was found to be more common in male population which may be attributed to their more outdoor activities than females. The male to female ratio was 7:3. The most common cutaneous site of rhinosporidiosis in our study is ocular region followed by parotid region, cheek, chin and extremities. In one of the patients multiple site involvement with recurrence was also noted.

Conclusion: Although the commonest site of rhinosporidiosis is mucosa, still in rare instances it can also present as cutaneous lesions. It is more common especially in backward and tribal areas due to lack of awareness, poverty and poor hygienic conditions. The disease responded well to wide local surgical excision and follow up treatment with Dapsone.

Keywords: Rhinosporidiosis, Cutaneous, Histopathology

Background

It is caused by Rhinosporidium seeberi, traditionally thought to be a fungus but actually an aquatic protistan parasite[1]. Rhinosporodiosis seeberi was initially considered a fungus of the sporozoan classification[2]. However, electron microscopic, histopathological, and molecular studies indicate that it is a eukaryote pathogen rather than a fungus[3]. It has been recently included into a new class known as Mesomycetozoea[4]. It is an endemic disease with more cases being reported from India and Sri Lanka[5].

Even though nose and nasopharynx are common sites there have been cases reported involving larynx, oropharynx, conjunctiva, lacrimal sac, trachea, bronchus, bone, vulva, vagina, penis, urethra, skin and subcutaneous tissue[6]. Rarely dissemination can occur with involvement of limbs, trunk, viscera and brain causing fatal outcome[7].

It is endemic in developing states like Chhattisgarh where it is more prevalent in tribal population. It can cause many symptoms depending upon the site of disease onset like nasal blockage, epistaxis, itching, rhinorrhoea. It can grow to advanced stages of disease progression if left untreated. Definite diagnosis is made by Histopathological examination. Treatment of choice is wide local excision and one year course of Dapsone to prevent further recurrence.

Material and Method

All the cases of cutaneous rhinosporidiosis which were observed in the past three years in tertiary medical health care centres of Chhattisgarh were included in this study. The cases which were suspected to be of cutaneous rhinosporidiosis based on radiological findings were included. As cutaneous rhinosporidiosis is a rare manifestation of rhinosporidium seeberi, the cases which were diagnosed as an incidental finding on histopathological examination were also included. The specimens were fixed for 24 hours before grossing. The specimens which showed autolytic changes were excluded. After fixing the specimens grossing was performed and the slides were stained with hematoxylin and eosin stain. The stained slides were examined under the microscope to establish the diagnosis of rhinosporidiosis.

Results

Here we are presenting a case series of ten rare cases of cutaneous rhinosporidiosis with unusual site of presentation. The age ranged from 2-6th decade. The incidence was found more in male population with male to female ratio being 7:3. Most of the cases were from remote tribal areas surrounding the district. The most common cutaneous site of rhinosporidiosis in our study is ocular region followed by parotid region, cheek, chin and extremities. One of the case of 61 year old male showed recurrence of rhinosporidiosis with presence of rhinosoridiosis at more than one location, sites being hand and thigh. There was no local rise in temperature and associated pain. The complete blood count showed eosinophilia in nearly all cases and raised erythrocyte sedimentation rate.

Table 1: Age and site distribution

S.no	Age	Sex	Site
1.	24	M	Cheek
2.	61	M	Hand and Thigh
3.	12	M	Eyelid
4.	14	F	Eyelid
5.	16	M	Palpebra
6.	60	F	Thigh

7.	25	F	Arm
8.	60	M	Chin
9.	36	M	Calf
10.	46	M	Parotid swelling

Case-Cheek rhinosporidiosis

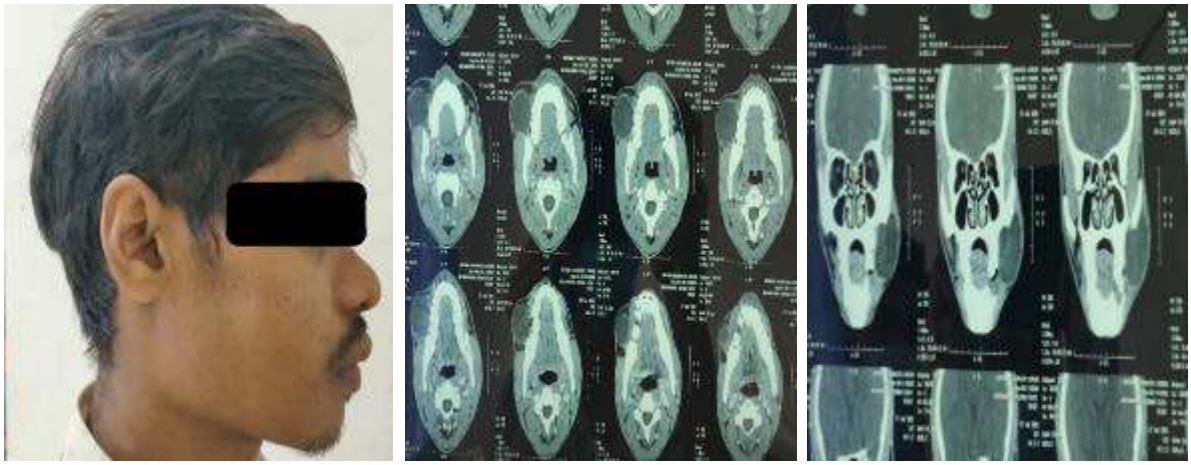


Figure 1: Ill-defined non tender cheek swelling; Figure 2, 3: Thick walled hypodense lesion with intact skin on left cheek

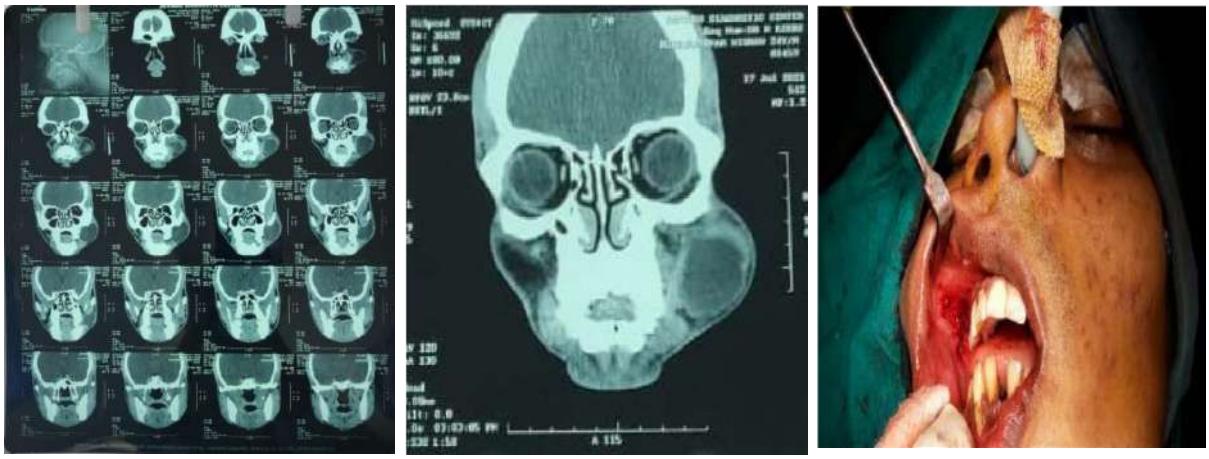


Figure 4,5: Well defined thick walled hypodense lesion; Figure 6: Intraoral burst open swellin with fluid discharge

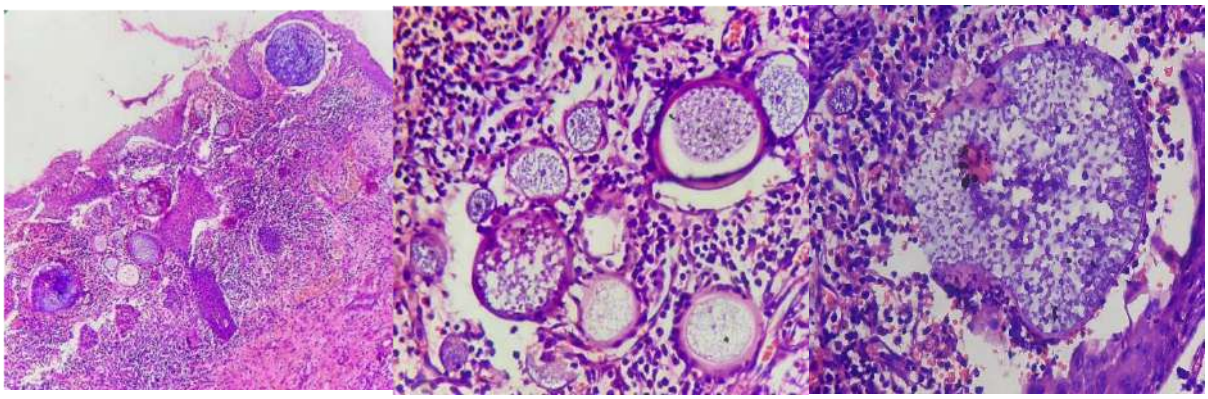


Figure 7: H&E(4x) Multiple sporangia with surrounding inflammation; Figure 8: H & E(10x) Mature sporangia with endospores; Figure 9: H&E (40x) Rupture of sporangia

with spillage of endospores

Case- Cheek rhinosporidiosis simulating parotid tumor

Figure 10: Ill defined parotid swelling; Figure 11: Right Parotid sialography showing collection of dye in cystic space

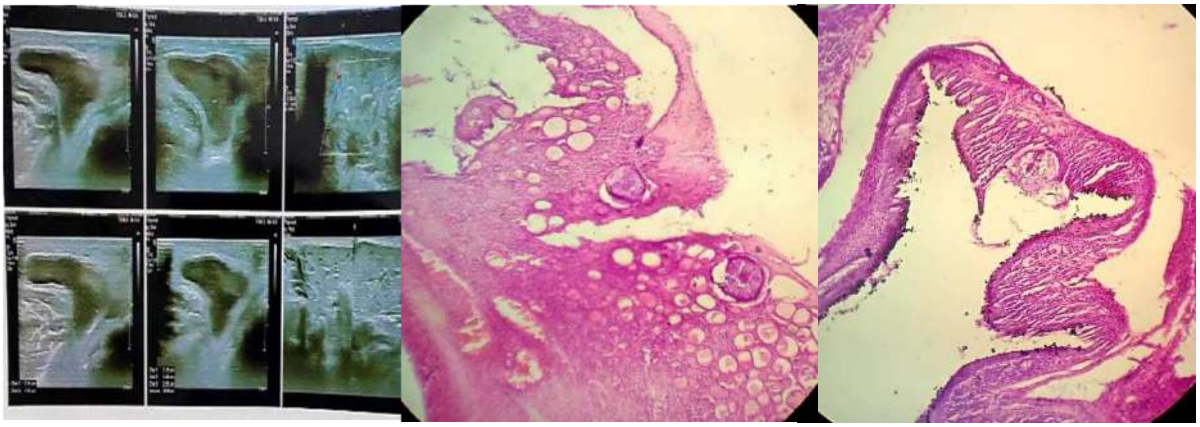


Figure 12: Ultrasoun show a hypoechoic anechoic cyst within Right Parotid gland; Figure 13,14: H & E(4x) Parotid tissue showing multiple sporangia of varying sizes

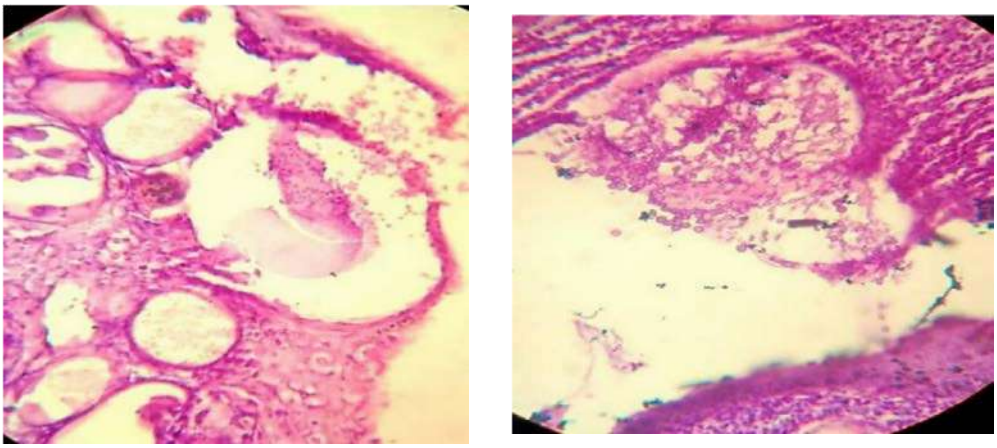
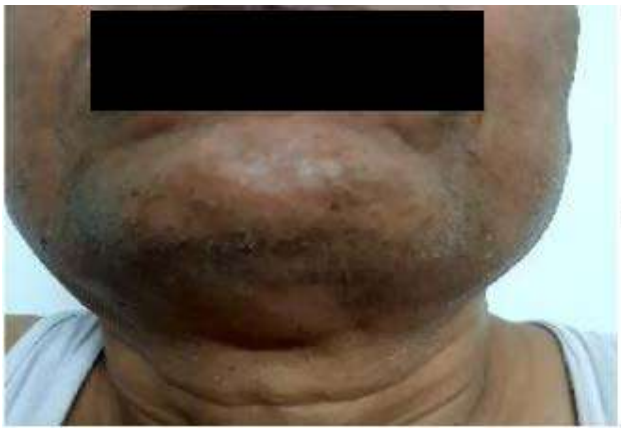


Figure 15: H&E(40x) Mature sporangia with endospores; Figure 16: H & E(40x)-Rupture of sporangia

Case- Chin rhinosporidiosis



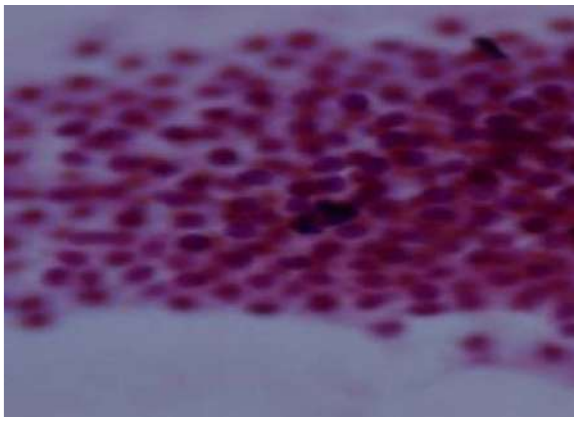
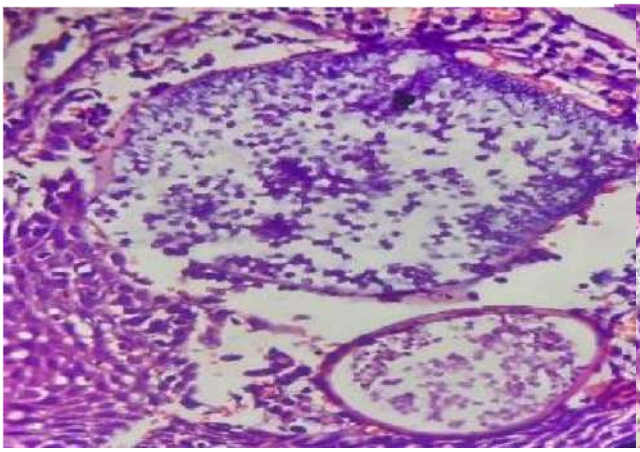


Figure 17: Ill-defined non tender chin swelling; Figure 18: H&E(40x) Multiple endospores



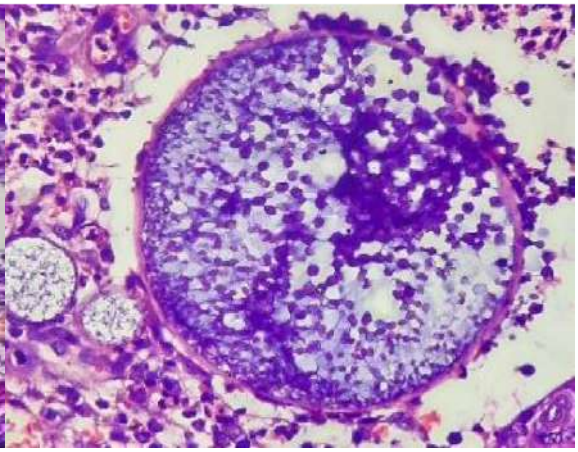
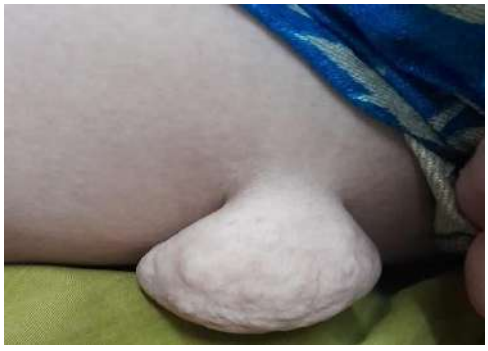


Figure 19: H&E(40x)- Ruptured sporangia; Figure 20: H&E(40x)- Intact sporangia with endospores

Case- Thigh rhinosporidiosis



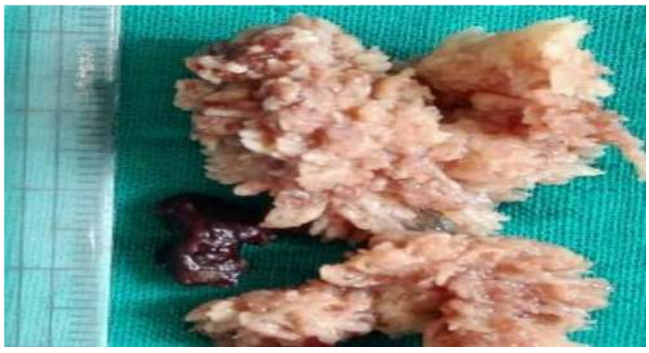


Figure 21: Pedunculated mass ove Right Thigh; Figure 22: Multiple grey white soft tissue bits

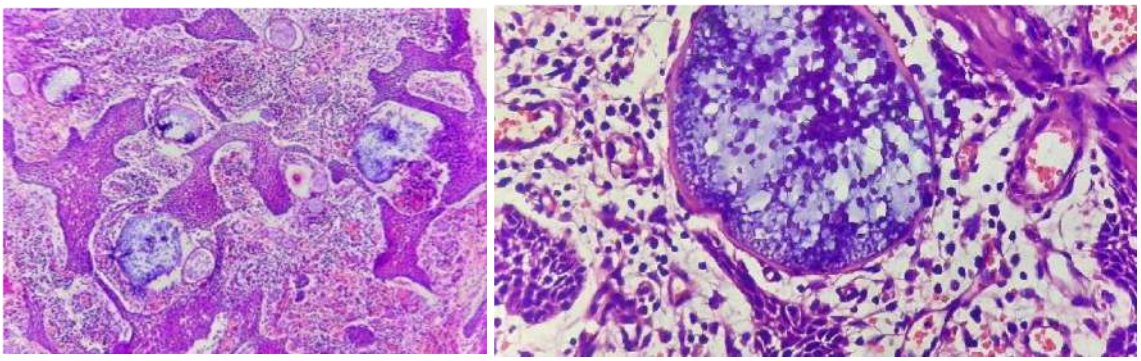


Figure 23: H&E(10x) Inflammatory cells surrounding spores; Figure 24: Intact mature sporangia

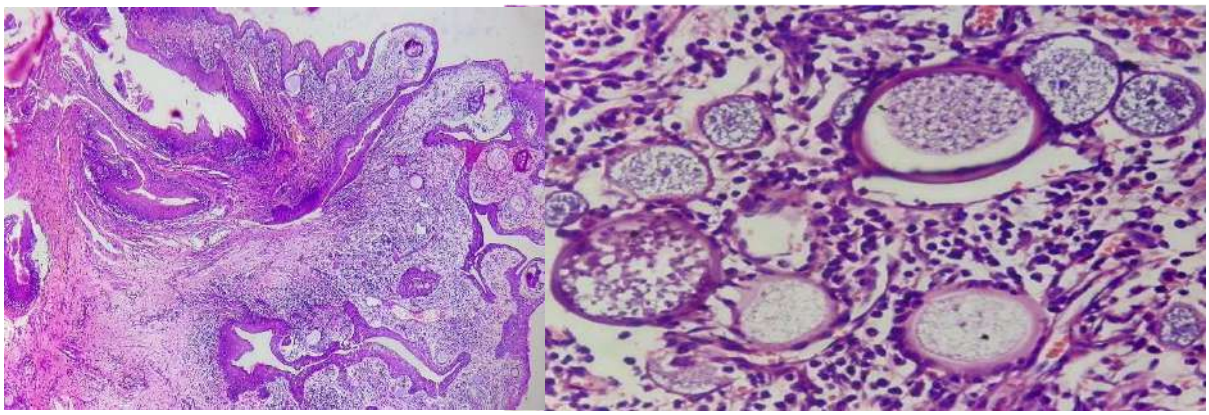


Figure 25: H&E(10x) Lid epithelium showing multiple sporangia;Figure 26: H&E(40x) Multiple sporangia with endospores

Discussion

Rhinosporidiosis has been reported from all over world, but the highest incidence has been from Sri Lanka and India. In India, Chhattisgarh is known to be an endemic area[8]. Rhinosporidiosis seeberi cannot be isolated in synthetic media in vitro, although it grows well in cell culture[9]. The final diagnosisis made by histopathological examination of slides made from the representative site.

Rhinosporidiosis has been seen associated with rural and aquatic sorroundings and it spreads via direct transmission of spores through dust, or wearing infected clothing, direct touching, swimming in infected water, trauma, followed by autoinoculation in some rare cases.

In our case series we found seven of the ten cases to be belonging to tribal community. The usual presentation of Rhinosporidosis is that of a fungating mass which bleeds on touch. But in our series, all the cases of cutaneous Rhinosporidiosis presented as soft tissue swellings, gradually increasing in size. The site are unususal site of presentation for Rhinosporidiosis i.e. ocular region, cheek, chin and extremities.

Cases includes a 24-year-old male from Korba district presented with chief complaints of swelling over right cheek for 2 months gradually increasing in size. There was negative history of pain, nasal blockage, discharge from nose or epistaxis. The swelling spontaneously burst open intraorally on the day of hospital admission causing draining of clear fluid inside the oral cavity (Fig 6). On examination a cystic swelling measuring 4.5x3.5x1.5cms over right buccal mucosa. Swelling was soft, non-tender and was not adherent to the underlying

structures(Fig1). It was followed by CT scan which showed well defined hypodensity seen around the crown of 2nd and last molar tooth at right side in mandible with breach in enamel region (Fig 2-5). On microscopy we found multiple sporangia of Rhinosporidum seeberi containing multiple spores (Fig 7-9). Kumar ES *et al*[10] have also documented a case report of rhinosporidium seeberi presenting as a cheek swelling on a 13 year old female child. Due to chronicity of the lesion, there was ruture of sporangia causing spillage of spores causing exacerbation of clinical symptoms (Fig 9).

Another case had near similar presentation where a 46 male, farmer by occupation presented to OPD with swelling in right cheek for 4 months and watering from right side of mouth for 4 months (Fig 10). On examination there was a unilateral painless swelling over right cheek which is insidious in onset for 4 months which is gradually progressive, increasing in size (Fig 11-12). Swelling increases while eating food but not associated with pain. There is history of watery discharge from right buccal mucosa which is watery in consistency, white in colour, non-blood stain since 4 months. Discharge was present only while eating food in scanty amount. Not associated with pain. There is negative history of facial asymmetry, trauma. Clinically a diagnosis of parotid duct cyst was made. On FNAC swelling decreased in size after aspiration and studied smears show abundant inflammatory cells predominantly of neutrophils, plasma cells and lymphocytes. Background shows necrotic cellular debris, macrophage and degenerated cells. A pathological diagnosis suggestive of acute on chronic inflammatory lesion was made. Surgery was performed and specimen was sent in pathology department which showed presence of multiple sporangia containing multiple spores (Fig13-16). A diagnosis of rhinosporidiosis seeberi was made. Sudarshan V *et al*[11] have also documented three cases of rhinosporidiosis of parotid duct presenting clinically as a parotid duct cyst. Rhinosporidiosis was diagnosed on histopathology.

A 12-year-old boy from urban area presented to eye opd with history of swelling over eye for 2 months. The swelling was painful, gradually increasing in size and simulated a tumor. The boy was attending swimming classes in local swimming pool. Due to his lesion over eye, he could no longer continue his swimming lessons and presented to eye opd of our medical college. On examination there was a swelling over left lid measuring 0.3x0.3x0.2cms which was tender on touch. Due to small size of the lesion and cosmetic reasons the lesion was excised in minor OT the very same day and eye patching was done. The soft tissue sample was sent for histopathological examination. On microscopy multiple spores of rhinosporidium seeberi were found (Fig 25-26). Sharma KD *et al*[12] has also documented ocular rhinosporidiosis simulating a tumor.Similar picture was present in two more children age being 14 year female and a 16 year male who had history of taking bath in community pond site being eyelid and palpebra.

In another incidence a 60-year-old male presented to the cytology department with swelling over chin. The swelling was 2x2x1cm soft, non-tender, gradually increasing in size (Fig 17). FNAC was done twice and yielded paucicellular material containing spore like material (Fig 18). Surgical excision was done and it showed presence of multiple sporangia of rhinosporidium seeberi containing multiple spores(Fig 19-20).

On separate occasion a 60 female presented a pedunculated mass over inner thigh measuring 2x1.5.1 cm present for 51 year. The mass was non tender, gradually increasing in size. Several passes were given but it didn’t yield any material except for blood and hemorrahge. Later on, she underwent surgery and her specimen was sent for histopathological

examination (Fig 22). Diagnosis of rhinisporidosis was made in histopathology.(Fig 23-24). Deshpande Ah *et al*[13] have also reported a case of polypoidal and warty skin growths on leg in a 28-year-old male which revealed numerous sporangia and spores of R. seeberi on microscopy.

Other cases include case of an elderly male patient from nearby village presented to the surgical department with swellings at hand and thigh. The swellings were ill defined measuring approximately 2x2x1cm, non-tender gradually increasing in size. Patient had outside FNAC report suggestive of lipoma. Surgery was done and specimen came to our department. We had received two grey, white soft tissue pieces measuring 2x2x1cm each. On microscopy it turned out to be a case of rhinosporidium seeberi. In this particular case there was presence of rhinosporidiosis at multiple sites and on further interrogation it was found that patient had mucosal rhinosporidiosis in past and had been operated for it. Prasad *et a* [14] has documented presence of R. Seeberi as multiple cutaneous lesions on malar aspect, infraorbital and supraorbital region, right shoulder and over the back near the tip of scapula. Oral cavity also revealed a polypoidal lesion, in the base of the tongue. Excision biopsy confirmed the lesions as disseminated cutaneous rhinosporidiosis. In rest of the three cases the patients had presented in surgery opd as soft tissue swelling in extremities. Surgical excision was done, and the cases were dignosed as rhinisporidiosis as an incidental finding. The cases were treated with wide local excision of the lesion followed by Dapsone. There was remarkable improvement in symptoms of the patients by giving post-surgical dose of Dapsone. Dapsone ceases the maturation of the spores and accentuation of granulomatous response[15].

Conclusion

Rhinosporidiosis is more common in backward and tribal population. Due to lack of awareness and health infrastructure in rural areas the patients report late, in advanced stages. The disease responded well to wide local surgical excision and follow up treatment with Dapsone.

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