Case Report

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Rhinosporidiosis of Parotid Duct Presenting as Consecutive Bilateral Facial Swelling: A Rare Case Report and Literature Review

Surgery Section

SANTANU SARKAR[1], SOUMYAJYOTI PANJA[2], ARGHYA BANDYOPADHYAY[3], SHREOSEE ROY[4], SANDEEP KUMAR[5]

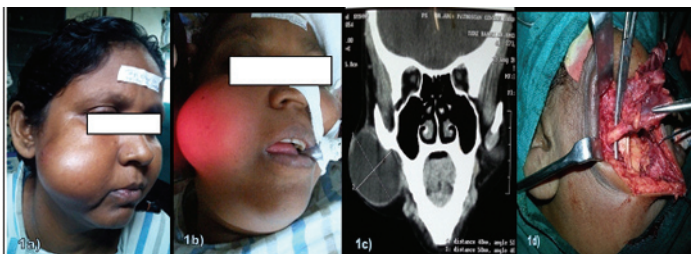
ABSTRACT

Rhinosporidiosis is a chronic granulomatous infection caused by *Rhinosporidium seeberi*. Sporadic cases of rhinosporidiosis has been reported from many countries but is endemic in Southern India (Madurai, Ramnad, Rajapalayam and Sivaganga), Nepal, Bangladesh and Sri Lanka. This disease commonly affects the mucous membrane of the nose or naso-pharynx and presents as a leafy, polypoidal mass. The reported extranasal sites include the oro-pharynx, eye, ear, larynx, trachea, bronchi, skin and genital mucosa. It may also become disseminated to present as a generalized form. In our case a 40-year-old female from rural West Bengal (Eastern India) presented with right sided facial swelling. Our provisional diagnosis was parotid duct cyst on the basis of careful history, scrupulous clinical examination and relevant investigations comprising CECT scan of face. Although Rhinosporidiosis was not taken into consideration in the clinical differential diagnosis, it was eventually diagnosed postoperatively by histopathological examination of surgical specimen. Two months later in follow-up, the same patient presented to us with left sided facial swelling. We managed the left facial swelling successfully with minimally invasive surgery and 100mg twice daily dapsone for 6 months. We present this case firstly because Rhinosporidiosis of parotid duct (stensen's duct) is a rare entity and secondly non-neoplastic cysts of the salivary glands are also uncommon and represent only 2-5% of all salivary gland lesions. Furthermore our case emphasizes that the clinicians should aware of this rare clinical entity specially in endemic areas, because minimally invasive techniques and medications might solve the problem while helping patients to avoid surgical excision.

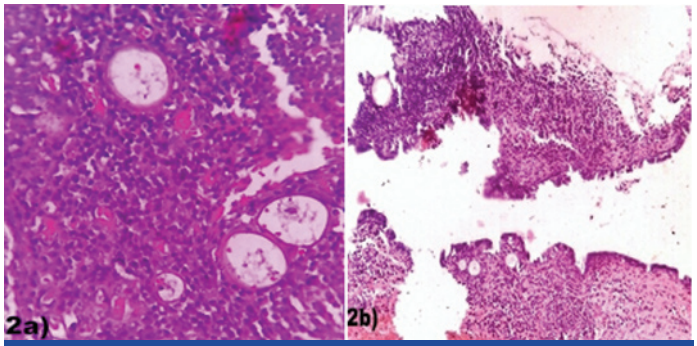
Keywords: Dapsone, Mesomycetozoea, Minimally invasive surgery, Stensen's duct cyst

CASE REPORT

A 40-year-old Bengali woman residing in the village of Galsi; Burdwan district of West Bengal (Eastern India) presented to our surgery outdoor with a swelling [Table/Fig-1a] in her right cheek on February' 2014. The swelling was about the size of a marble ball when she first noticed it 3-months back. It gradually enlarged in size. She had experienced increase in the size of facial swelling while having meals along with mild pain. She gave history of consumption of unprocessed well water and also used to take bath in the pond. On extraoral examination, a soft, fluctuant and brilliantly transilluminant [Table/Fig-1b] swelling was seen measuring about 6×5cm on the right side of her face. The swelling was mildly tender on palpation, it's surface was smooth. The swelling was not fixed to the skin or underlying structures and the skin over the swelling appeared glossy without any rise of local temperature. Intraoral examination showed copious effusion of non-viscous serous fluid from the inflamed and stenosed stensen's duct opening while compressing the swelling extraorally. There was no cervical lymphadenopathy. Examination of ear, nose, nasopharynx and systemic examination did not reveal any other abnormalities. Routine laboratory investigations were within normal limits. Contrast Enhanced CT scan (CECT) of the face showed a thick-walled peripherally enhancing lesion overlying right masseter muscle measuring 5 X 4 cm [Table/Fig-1c]. Provisional diagnosis of parotid duct cyst was made from the aforesaid clinical and radiological picture and she had undergone superficial parotidectomy along with excision of parotid duct cyst [Table/Fig-1d]. Postoperative period was uneventful and she was discharged home on 6th post-op day. Diagnosis in the present case was established by histopathology, which revealed that the dilated duct of the parotid gland lined by columnar epithelium with sub-epithelial dense lympho-plasmacytic infiltration admixed with eosinophils and sporangia of varying sizes containing numerous daughter spores of *Rhinosporidium seeberi* [Table/Fig-2a,b]. Two months later in the follow-up visit, she presented to us with left sided facial swelling having similar clinical characteristics like right sided facial swelling [Table/Fig-3a]. We incised the stenosed intraoral opening of left parotid duct and cannulated the duct and the cyst with a properly sized no.10 infant feeding tube and sutured it to oral mucosa to keep it in position [Table/Fig-3b]. In that way, intraoral drainage of left parotid gland secretion was maintained



[Table/Fig-1]: (a) Front view of the patient showing right sided facial swelling (b) the swelling was brilliantly transilluminant (c) CECT of the face showing a thick-walled peripherally enhancing cystic lesion overlying right masseter muscle (d) Intraoperative picture during excision of cyst(parotid duct hooked by a artery forcep).



[Table/Fig-2]: (a) Sporangia containing numerous daughter spores (H&E stain, X 100) (b) Lining of the parotid duct showing heavy inflammatory reaction with spores of Rhinosporidiosis (H&E stain, X 40).

[Table/Fig-3]: (a) Front view of the same patient showing left sided facial swelling .(b) Intraoral picture showing stensen's duct stent in-situ(white vertical arrow).(c) Recent photograph of the patient after 12 months follow-up.

(internal fistulation). We removed the stent after 2-weeks and daily 200mg dapsone was prescribed for next 6-months [Table/Fig-3]. We requested her to avoid the use of unprocessed well and pond water and advised her to use deep tube well water which is available in their village for public use. In last 16-months of follow-up period there was no sign of recurrence.

DISCUSSION

Rhinosporidiosis is a chronic granulomatous infection, predominantly affecting the loosely attached mucous membrane of both humans and animals. Previously the causative organism has been considered to be a fungus but now it has been included into a novel group of fish parasites mesomycetozoea (a class of microorganisms intermediate between animals and fungi) based on phylogenetic analysis of *Rhinosporidium seeberi*'s 18S Small-Subunit Ribosomal DNA [1,2]. Transepithelial infection through the traumatised epithelium (most commonly through nasal mucosa) is the usual mode of infection from the natural aquatic habitat of this organism [3]. Majority of the cases occur in upper respiratory tract, notably anterior nares, nasopharynx, larynx, and soft palate. It usually presents as a polypoidal strawberry like mass, pink to purple in colour. It is a highly vascular lesion which bleeds easily on touch. Rhinosporidiosis can occur at any age but commonly encountered between 20 to 35 years, males are four times more commonly affected when compared to females. Present case of rhinosporidiosis of the parotid duct may be due to washing mouth and taking bath using infected pond and well water. The natural patulous opening of the parotid duct in the oral cavity could have promoted a quick passage of the spores into the parotid duct causing ductal stenosis and resulting in the development of the cyst.

The first case of rhinosporidiosis of the parotid duct was reported by Topazian RG in the year 1966 [4]. Involvement of the stensen's duct is rare and only few cases reported in the literature in last 10-years [5-10]. In most of the the published case reports, patients were presented with a cystic swelling in the cheek and were provisionally diagnosed as stensen's duct cyst [4,5-12]. Rhinosporidiosis was not suspected in any of these cases. The diagnosis was made by histopathological examination of the excised tissue. The mature stage of *Rhinosporidium seeberi* consists of characteristic large, thick-walled spherical structures with an apical pore called 'sporangia' (50-1000 µm) in various stages of maturation containing smaller daughter cells called 'sporangiospores' (20-80 µm). The sporangia and sporangiospores of the organism can be seen with typical fungal stains such as Gomori Methenamine Silver(GMS), Periodic Acid–Schiff (PAS), and mucicarmine as well as with standard haematoxylin and eosin stain. A purulent neutrophilic inflammation is common in areas where sporangia have ruptured [13]. The morphologic characteristics of *Coccidioides immitis* are very similar to *Rhinosporidium seeberi.* Sporangial endospores of *R.seeberi* are more numerous and larger in size in comparison to *C. immitis* on haematoxylin and eosin stain and *C. immitis* does not stain with mucicarmine [6].

Salivary duct cysts are true cysts. It's incidence is variable in different parts of the world. According to European and American literature, salivary duct cysts constitute approximately 10% of all salivary cysts [14]. On the other hand it is rare in Japan. Only 3 cases (0.5%) of salivary duct cyst was identified among 586 patients with salivary gland cysts at the Clinical Pathology division, Iwate Medical University Hospital, Japan between 1975 and 1999 [15]. An extensive search of English medical databases including pubmed; pubmed central and google scholar using keyword non-neoplastic salivary duct cyst revealed few interesting isolated case reports from India [14,16]. A retrospective study of 150 patients with salivary gland disease in the institute of medical sciences; Banaras Hindu University, Varanasi, India between 1997 and 2001 revealed that only 17(11.33%) patients were presented with non-neoplastic salivary gland disease involving the parotid gland. Parotid abscess was the most common cause followed by parotid cysts, which were seen in 5-cases without any case of parotid duct cyst [17]. Imaging studies including ultrasonography, CECT, MRI scan and Sialography plays an important role in diagnosis of salivary duct cysts.

Surgical excision is the treatment of choice in a case of parotid duct rhinosporidiosis, though minimally invasive surgery along with dapsone can be tried in selected cases [18]. Treatment with the drug dapsone appears to be promising which appears to arrest the maturation of the sporangia and to promote fibrosis in the stroma, when used as an adjunct to surgery [19]. Recurrences are known to occur due to spillage of endospores on the adjacent tissue or incomplete excision.

CONCLUSION

In conclusion we emphasizes that clinicians should aware of this rarely encountered entity because minimally invasive techniques and medications might solve the problem while helping patients to avoid surgical excision. Furthermore; diagnosis of rhinosporidiosis should be kept in mind during histopathological examination of any granulomatous lesions involving the mucous membranes especially in the endemic areas.

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PARTICULARS OF CONTRIBUTORS:
1. Assistant Professor, Department of General Surgery, Burdwan Medical College & Hospital, West Bengal, India.
2. PG Resident, Department of General Surgery, Burdwan Medical College & Hospital, West Bengal, India.
3. Assistant Professor, Department of Pathology, Burdwan Medical College & Hospital, West Bengal, India.
4. Demonstrator, Department of Pathology, Burdwan Medical College & Hospital, West Bengal, India.
5. PG Resident, Department of General Surgery, Burdwan Medical College & Hospital, West Bengal, India.

NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR:
Dr. Santanu Sarkar,
1/3/1 C Ramlal agarwala Lane, Sinthee, Kolkata-700050, West Bengal, India.
E-mail: drsantanu.surg@gmail.com

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