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Original Research Paper ENT

UNUSUAL CASE OF PAROTID DUCT RHINOSPORIDIOSIS: A CASE REPORT

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ABSTRACT Sialectasis is an uncommon condition characterised by abnormal dilation of salivary ducts. It can be due to sialoliths, stricture, recurrent infection or trauma. We encountered a case of parotid sialectasia left cheek. Complete excision was done. Biopsy confirmed as parotid duct rhinosporidiosis. Isolated involvement of parotid duct is very unusual. Hence we are presenting a case report of parotid duct rhinosporidiosis. Rhinosporidiosis should be included as one of the cause for sialectasia.

KEYWORDS : Rhinosporidiosis; Parotid duct; Sialectasia; Ductocele

INTRODUCTION

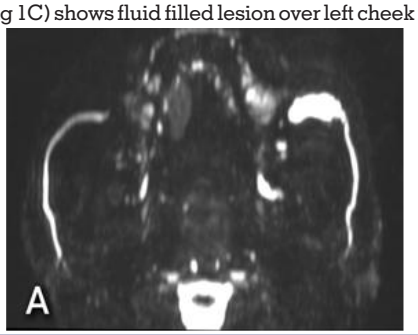
Sialectasis is cystic dilatation of the ducts of salivary gland due to ductal stricture or ductal calculi. Rhinosporidiosis is the unknown cause of sialectasia and has not been mentioned in literature. Occurrence of parotid duct rhinosporidiosis is very rare and few cases have been reported. It presents with soft cystic swelling over cheek. Rhinosporidiosis, a chronic granulomatous infective disease is caused by a cynobacterium, Microcystis aeruginosa. [1] It is endemic in South Asia being mainly reported from southern India and Sri Lanka. It typically involves mucous membrane of nose and nasopharynx and less commonly other organs. Final diagnosis could be made only after histopathology examination.

Case Report

A 41 year old female patient presented with swelling of left cheek of 2 months duration. Swelling is insidious in onset and gradually progressive in size. On clinical examination 3 x 2 cm size, cystic firm swelling was observed at left cheek region. Swelling was compressible. On intraoral examination multiple wound present at left buccal region due to previous attempt of parotid duct dilatation. Rest of the ENT examinations were normal.

On MRI neck examination T1 hypointense and T2 hperintense collection measuring 30 x 21 mm size seen in left buccal mucosa region. Posteriorly collection is closely abutting masseter muscle. Lesion is seen between left zygomatic muscle and masseter muscle probably involving left stensen's duct. No intraductal calculus was noted. Left parotid gland dilated. Features likely represented abscess.

On MRI Left parotid sialogram well defined fluid collection observed over left side face along the anterior aspect of the masseter muscle. The collection measures 8 x 21 x 9.5 mm and appears isointense on T1 and hyperintense on T2 images and shows peripheral enhancement on post contrast images. On thin T2wt images the collection is communicating with the left parotid duct and shows few intermediate to hypointense filling defects within it suggestive of left parotid duct sialocele (Fig 1A). MRI coronal STIR image (Fig 1B) and 3D volume rendered image (Fig 1C) shows fluid filled lesion over left cheek region.



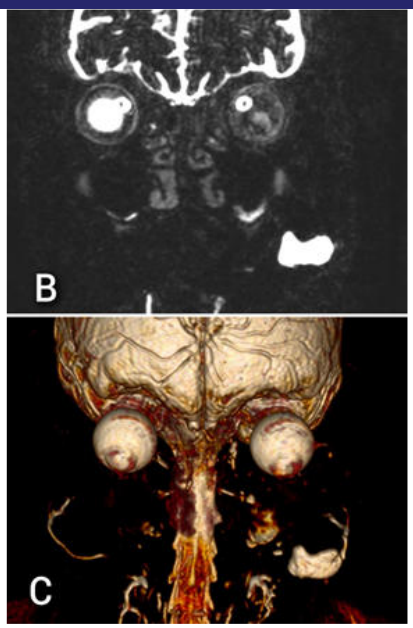
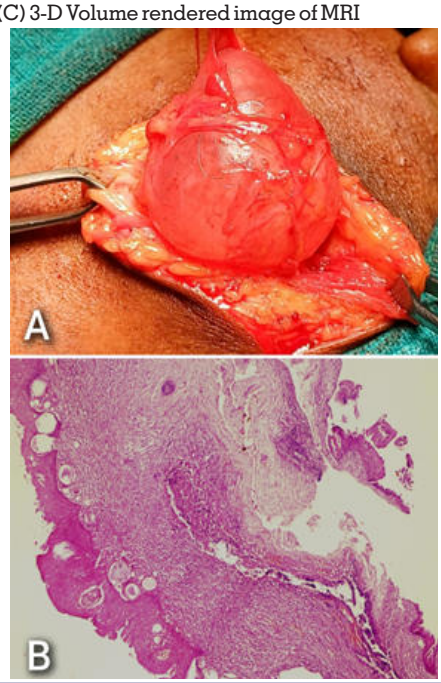


Figure 1: (A) On MRI Sialogram thin T2Wt image the collection is communicating with the left parotid duct with few intermediate to hypointense filling defects within it. (B) On MRI Coronal STIR image collection appears hyperintense over left cheek. (C) 3-D Volume rendered image of MRI



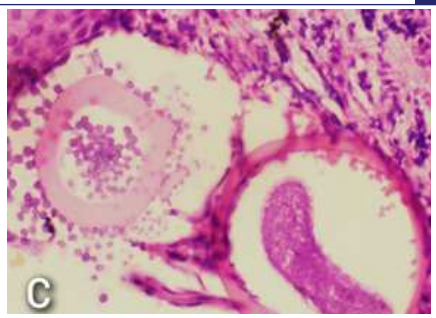


Fig 2: (A) Intraop Picture of left parotid ductocele. (B) Photomicrograph showing thick walled sporangia containing endospores (H&E 40x). (C) High power view of sporangium and endospores (H & E 400x)

Extraoral excision of left parotid ductocele and ligation of duct was done under general anesthesia (Fig 2A). On histopathology parotid duct was dilated with metaplastic stratified squamous epithelium lining the cyst with presence of numerous thick walled sporangia containing the endospores which confirms the rhinosporidiosis of parotid duct (Fig 2B & 2C). Detailed examination of nose and nasopahrynx was done for the search of rhinosporidiosis lesion, but lesion was not found at other site.

DISCUSSION

First case of parotid duct rhinosporidiosis was reported by Mahadevan R in 1952.[2] Thereafter Karunaratne (1964) reported a case. [3]Sialectasis is the unusual dilation of a salivary duct which occur as a result of chronic obstruction secondary to sialoliths. Strictures, stenosis, masseter hypertrophy and chronic inflammation of duct are other reasons.[4] Involvement of parotid duct by rhinosporidiosis could be also one of the cause for parotid duct dilatation. Rhinosporidiosis is endemic in Odisha, Chhattisgarh, Bengal and few parts of South India. Rhinosporidiosis most commonly involves nose and nasopharynx. Other body parts are rarely involved. Isolated involvement of parotid duct is very rare finding. Very few cases have been reported in world literature.

Patient presents mainly with soft, cystic and compressible swelling over cheek externally which increases during meals. The patient rarely complains about pain and discomfort. The presumed mode of infection from the natural aquatic habitat of the organism is through the traumatized epithelium. One study suggests that possibility of natural patulous and everted anatomy of the orifice of the parotid duct inside the oral cavity may promote a quick passage of the spores into the duct during washing the mouth cavity in pond water leads to development of the disease in parotid duct.[5]

Sialography, CT scan and MRI are considered to be the imaging modalities for diagnosis of dilatation of salivary ducts. Sialography shows dilatation of the salivary ducts with a well-defined border. CT scan can demonstrate the presence or the absence of a sialolith. MRI is the most effective method demonstrating location and degree of stricture or dilatation. MRI is the gold standard for exploring salivary glands. Indeed, MRI and sialo-MRI showed ductal ectasia with salivary retention and cystic mass.

Standard operative procedure for parotid ductocele is surgical excision with ligation of duct. Various modification and surgical approaches have been tried. Ductocele can be approached by intraoral or extraoral technique. Local and general anesthesias both are equally safer and depend upon size and location of lesion.

Several treatment options are available for sialectasia. The choice of therapeutic modality depends on the size, the location of dilatation and the surgeon's experience. Conservative methods like repeated aspiration and compression, dilation of the papilla and stent placement may be performed but usually associated with recurrence. Intraglandular ductocele may require superficial parotidectomy to assess the complete mass. Marsupialisation for distal sialectasis, distal dilatation of salivary duct, excision of the dilated portion of the duct, reimplantation into buccal mucosa and excision of the Stensen's duct are the few surgical method mentioned in literature. End to end anastomosis and recannulation of duct has also been tried.[6] Superficial parotidectomy was also performed in few cases.[7,8] Currently, advances in minimal invasive sialoendoscopy technique has reduced the use of aggressive surgical treatment for sialectasia.

Imaging and cytological study could not help in the diagnosis of rhinosporidiosis if it involves very unusual sites. Histopathology test remains gold standard for the confirmation of rhinosporidiosis. Histopathological features that are readily recognizable in haematoxylin and eosin-stained sections shows sporangia (50-1000 μm) develop from individual spores into small uninucleate cysts that enlarge and develop a chitinous eosinophillic wall. The sporangia and sporangiospores can also be visualized with fungal stains such as Gomori methenamine silver and PAS, and mucicarmine.[9] Wet mount and routine cytological stains can act as an adjunct to histopathological examination as a simple tool for definite diagnosis of this condition.

Differential diagnosis in these cases were sialocele, parotid cyst, parotid duct cyst, ductocele, retention cyst, abscess, granulomatous lesion, mucous retention cyst and minor salivary gland neoplasm and need to be differentiated. Rhinosporidiosis could also be one of the causative factor for formation of ductocoel and should always be looked for in histopathology of specimen to rule out Rhinosporidiosis.[10]

CONCLUSION

Isolated Parotid duct rhinosporidiosis is uncommon finding. Rhinosporidiosis need to be included as one of the causative factor for sialectasis particularly in endemic zones. Routine cytological stains with histopathological study will aid in definitive diagnosis of rhinosporidiosis at not so usual sites.

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