

Case Series

Rare cases encountered in otolaryngology and head neck surgery

Tabish Maqbool*, Kulvinder Singh Mehta, Showkat Ahmed Showkat, Rauf Ahmed

Department of Otolaryngology and Head Neck Surgery, GMC Srinagar, Jammu and Kashmir, India

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*Correspondence:

Dr. Tabish Maqbool,

E-mail: Tabish.maqbool31@gmail.com

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ABSTRACT

In our day to day otolaryngological practice we encounter many cases, some of which are very rare and worth reporting. We report very rare three cases seen during our practice: presence of tooth in anterior ethamoids causing chronic sinusitis, and NK/T Cell lymphoma (nasal variant) and An accessory submandibular gland. There have been few case reports which mentions about the presence of tooth in maxillary sinus leading to sinusitis but none mentions of presence of the tooth in anterior ethamoids leading to sinusitis. Extranodal NK/T cell lymphoma, nasal variant is a rare and aggressive type of non-Hodgkin's lymphoma that develops outside the lymphatic system especially in the nose. Accessory submandibular gland is extremely rare. This is the fifth case reported so far worldwide.

Keywords: Submandibular, Sinusitis, Lymphoma

INTRODUCTION

Ectopic presence of tooth in nose causing sinusitis is also a very unusual phenomenon. Supernumerary teeth are usually present in 0.1-1% of population.¹ NK T cell lymphoma is very infrequently seen malignancy of nasal cavity. Accessory salivary gland tissue refers to lobules of salivary gland tissue which are completely separate from the main gland at a variable distance. Accessory submandibular gland is extremely rare condition.

CASE SERIES

Case 1

Presence of ectopic tooth in anterior ethmoidal cells

50 year old male with no comorbidities presented with history of nasal obstruction and headache since last 7 months. Patient had taken some over the counter medications for it but was not relieved and presented to our department with the same complaints. On examination

Tenderness was present in ethamoids. Patient was diagnosed with chronic rhinosinusitis and was put on inhalational steroids for a period of three weeks.



Figure 2: Mucosal thickening and hyper dense tooth present in anterior ethamoids (coronal view).

After three weeks Non contrast tomography of nose and para nasal sinuses was done which showed the presence of hyper intense dense substance in anterior ethamoids on left

side and mucosal thickening of bilateral ethamoids sinuses, left greater than right.

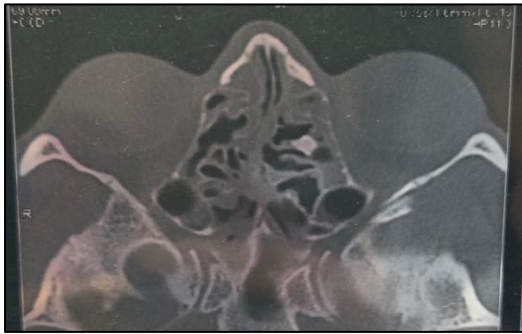


Figure 2: Mucosal thickening and hyper dense tooth present in anterior ethamoids (Saggital view).



Figure 3: Tooth removed after FESS.



Figure 4: Tooth removed after FESS.

Oral cavity showed normal dentition. Patient underwent functional endoscopic sinus surgery and during the procedure a tooth was found and removed from the anterior ethamoids on left side. After surgery symptoms of the patient completely resolved and no recurrence was seen in the patient who was on regular follow-up for two years.

Case 2

NK/T cell lymphoma (nasal variant)

55 year old male presented to us with complaints of crusting of nose for which he used to take some over the counter medications but was not relieved. Patient then started with facial swelling and came to our department.

Diagnostic nasal endoscopy was done which showed extensive bilateral crusting in both nasal cavities with ulcero proliferative lesion involving left side of septum causing septal perforation and going to the opposite side.



Figure 5: Preoperative CT scan findings showing mass in the left nasal cavity.



Figure 6: Preoperative CT scan findings showing mass in the left nasal cavity.



Figure 7: Post treatment pictures of patient presenting with defect.



Figure 8: Post treatment pictures of patient presenting with defect.



Figure 9: Post treatment pictures of patient presenting with defect.

Biopsy was taken which came out to be inconclusive. MRI was advised which showed diffuse enhancing soft tissue thickening involving forehead and dorsum of nose with dehiscent septum and dehiscent right nasal bone with right lateral rectus palsy. Biopsy of the nasal mass was again taken which showed features of NK/T cell lymphoma stage 1E complicated with superadded skin infection. Immunohistochemistry, CD3, CD56, CD8, CD 40RO confirmed NK/T cell lymphoma. Patient was planned for chemoradiotherapy and received DeVIC protocol (Dexamethasone, Etoposide, ifosfamide, and carboplatin) with subsequent radiotherapy. During the process of treatment post chemotherapy patient started with defect in the right nasal wall. After 8 weeks of completion of treatment patient was advised PET scan on follow up which showed no signs of recurrent or residual disease.

Extra nodal NK/T cell lymphoma, nasal variant is a rare and aggressive type of non-Hodgkin's lymphoma that develops outside the lymphatic system especially in the nose. It develops from NK cells and cytotoxic T cells. Prescilla b wood in 2011 reported a case of NK T cell lymphoma nasal type which was seen in a 30 year old male.³ This patient underwent chemotherapy (CHOP regimen) and subsequent radiotherapy. Tababi reported 15 cases of extranodal NK T cell lymphoma nasal type from 1990 to 2009. 74% of patients were having stage IE.⁴ Radiotherapy alone was performed for four patients with stage IE disease. Combined treatment (radiotherapy and chemotherapy) was administered to four patients with diffuse stage IE, while cyclophosphamide, doxorubicin, vincristine, prednisolone (CHOP) chemotherapy alone was administered in one patient with diffuse stage IE disease. But this study does not give the conclusion based on the outcome of patients depending on the treatment. The treatment modalities of extranodal NK T cell lymphoma is complex. Some authors are not in the favour of surgery and chemoradiation remains the main modality of treatment.

Case 3

Accessory submandibular gland

22 year old female with no comorbidities presented with pain in right submandibular area since last 3 months. On examination submandibular region was seen to be tender. Oral examination was normal. Oral digital palpation was also normal. Ultrasonography was done which showed chronic sialadenitis. Orthopantogram showed a stone in the duct. Patient was taken for submandibular gland excision. On table, an incidental finding of an unusual J shaped tissue was found at the level II a lymph nodes close to the enlarged nodes. This tissue was also found to have a duct of its own. This tissue was removed and duct ligated along with the normally placed submandibular gland. Both the specimens were sent for histopathology in separate biopsy containers. The biopsy revealed that this tissue was a seromucinous salivary gland tissue and thus confirmed the presence of accessory submandibular gland.



Figure 10: preoperative picture showing sialolithiasis.

DISCUSSION

Supernumerary teeth is usually present in 0.1-1% of population.¹ Nasal tooth results from ectopic eruption of supernumerary teeth. The diagnosis can be made on the basis of clinical and radiological findings. The presence of tooth in nasal cavity is a rare finding and even rarer is the presence of tooth in nasal cavity leading to sinusitis. There have been few case reports which mentions about the presence of tooth in maxillary sinus leading to sinusitis but none mentions of presence of the tooth in anterior ethamoids leading to sinusitis.

Kayabasoglu et al reported a case in which a tooth was found in the maxillary sinus which led to the chronic sinusitis in a 46 year old female which was removed by Caldwell-Luc procedure.²

Accessory submandibular gland is extremely rare. This is the fifth case reported so far world wide. The first case was reported in 2000.⁵ In 2007 it was also recognised by Gadodia et al in a 20 year old male on magnetic resonance sialography.⁶ Bryan in 2013 reported a case where he found the presence of pleomorphic adenoma in Accessory

submandibular gland.⁷ Barrueco et al reported a 39-year-old female with symptoms of left submandibular sialoadenitis and sialolithiasis within the left accessory submandibular gland by magnetic resonance sialography (MR-Si).⁸ Togni et al reported that only 4 cases were detected till then.⁹

CONCLUSION

The possible cause of sinusitis is irritation of the surrounding mucosa, leading to the increase in the inflammation and inflammatory mediators which further increase the inflammation of the whole of the surrounding mucosa and this vicious cycle leading to sinusitis.

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