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Case Report
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Mandibular Respiratory Cysts Following Orthognathic Surgery: 2 Rare Case Reports

Nazlie Syyed[1], Rebecca Mason[2], Ewen Thomson[2] and Jeff Downie[2]*

[1]*Department of Oral and Maxillofacial Surgery, Queen Eilzabeth University Hospital, UK*

[2]*Department of Oral and Maxillofacial Surgery, Forth Valley Royal Hospital, UK*

Introduction

Mandibular cysts composed exclusively of respiratory epithelium are extremely rare. These benign cysts are thought to be iatrogenic in nature and attributed to entrapment of sinus or respiratory mucosa during the operative period. They can be known by a number of different names, such as respiratory mucoceles, respiratory implantation cysts, surgical ciliated cysts and surgical implantation cysts [1].

Respiratory cysts, or surgical ciliated cysts as first described by Kubo in 1927 occurred in the maxilla following surgical treatment of maxillary sinusitis [2]. The majority of cases has been described in Japanese literature and is a common maxillary cyst, detected in up to 20% of patients following radical maxillary sinus surgery [3]. Cases following orthognathic surgery in the mandible are rarely described in the literature. When diagnosing these cysts, a number of differential diagnoses should be considered including inflammatory or neoplastic processes. We present 2 cases of mandibular respiratory cysts more than 10 years post orthognathic surgery.

Case Reports

Case 1

A 38 year old male was referred by the Public dental service in 2015 for management of an upper right 6 which was in close proximity to the maxillary sinus. During the initial investigation, an incidental finding of a large radiolucency was noted in the symphyseal region of the mandible. When questioned, the patient denied any symptoms arising from the anterior mandible.

The patient had undergone a segmental Le Fort 1 osteotomy and a Bilateral Sagittal Split Osteotomy (BSSO) with an augmentation genioplasty 18 years prior with no complications and an unremarkable recovery.

Initial investigations included an Orthopantogram (OPT) to assess the right maxilla and this was subsequently followed by a CT scan of the mandible to further assess the extent of the cystic lesion. Both demonstrated a multiloculated cystic lesion with mild expansion of the labial cortex. Screws from the genioplasty plate were seen to be communicating with the cyst which extended from the lower right 6 to the lower left 6 of the mandible with a total diameter of 6 cm (Figure 1).

The clinical presentation was unremarkable, with no obvious bony expansion, no sensory deficit associated with the inferior dental nerve sensation and all associated dentition were noted to be vital. Due to the size of the cyst a biopsy was initially performed under General Anaesthetic and the previous genioplasty plates were removed simultaneously.

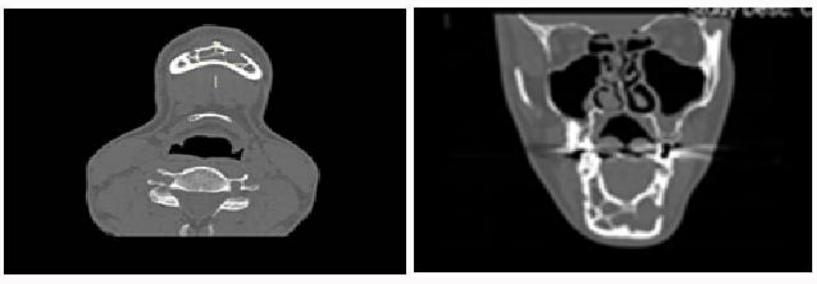

Figure 1: Pre op CT scan demonstrating the multilocular cyst associated with the genioplasty plate and screws.

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Correspondence:
Nazlie Syyed, Department of Oral and Maxillofacial Surgery, Monklands Hospital, Monkscourt Avenue, Airdrie, ML6 0JS; 2 StR Oral and Maxillofacial Unit, Southern General Hospital, Glasgow, G51 4TF, UK, E-mail: nazlie.syyed@doctors.org.uk

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Table 1: Differential Diagnosis.

Unilocular	Multilocular	Ill Defined Margins
Odontogenic Keratocyst	Ameloblastoma	Osteomyelitis
Residual Cyst	Odontogenic Keratocyst	MRONJ/ORN
Dentigerous Cyst	Giant Cell Granuloma	Fibrous dysplasia
Apical Granuloma	Odontogenic Myxoma	Cemento-osseus Dysplasia
Solitary Bone Cyst	Aneurysmal Bone Cyst	Multiple myeloma
Adenomatoid Odontogenic Tumour	Solitary Bone Cyst	Metastatic disease
Respiratory implant Cyst	Calcifying Epithelium Odontogenic Tumour	Primary bone Neoplasm
Cemento-Ossifying Fibroma	Odontogenic Fibroma	Bony invasion of Tumour
Langerhans Cells Histocytosis	Clear cell Odontogenic Tumour	Sarcoma
Radicular cyst	Calcifying Odontogenic Cyst	Pagets Disease
Lateral Periodontal Cyst	Respiratory Impant Cyst	
Ameloblastoma	Haemangioma	
Giant cell Granuloma	Mucoepidermoid Carcinoma	
Aneurysmal Bone cyst		
Clacifying Odontogenic Cyst		
Odontogenic Fibroma		
Haemangioma		

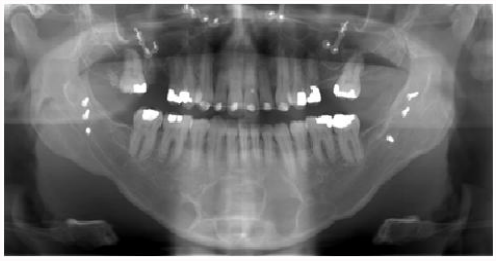

Figure 2: OPT post enucleation demonstrating bony infill.

The pathology demonstrated a thin cyst wall lined by a single layer of bland cuboidal respiratory type epithelium with an overall appearance of a mandibular respiratory cyst and the patient subsequently a second procedure to enucleate the cyst 5 months later. His recovery was unremarkable with bony infill demonstrated during follow up (Figure 2).

Case 2

A 25 year old female presented to the clinic complaining of a 2 week swelling associated with the anterior mandible which decreased in size with a one week course of oral metronidazole and Penicillin V. Her past medical history was unremarkable and she has previously undergone a Le Fort 1 osteotomy, BSSO and a genioplasty 10 years prior to this episode. No clinical notes were available.

Clinical examination demonstrated a firm, tender swelling in the lower labial sulcus with no extra oral swelling, and was described as having decreased in size following the antibiotic regime. Poor oral hygiene and periodontal disease throughout the oral cavity was also demonstrated, being more severe surrounding the lower anterior teeth. Vitality testing confirmed all teeth were vital.

An OPT revealed a unilocular cystic lesion in the midline of the mandible associated with the roots of the LR2 -LL2 and the screw tips of the genioplasty plate (Figure 3).

The cyst was enucleated with removal of the plate and screws

Figure 3: Pre op OPT demonstrating a unilocular cyst in the anterior mandible with the associated genioplasty plates and screws.

Figure 4: Post op OPT post operative demonstrating bony infill and healing.

under general anaesthetic. Pathology demonstrated a fully enucleated respiratory cyst. The patient is still under review but up until now her recovery has been unremarkable (Figure 4).

Discussion

Respiratory cysts located in the mandible are extremely rare and has been described in the literature on a handful of occasions. It is thought to be iatrogenic in nature following implantation of sinus mucosa during surgical procedures. In the cases mentioned above, the surgeries involved a le fort 1 osteotomy with a genioplasty augmentation. It is unclear if bone grafting occurred at this time. The same blade is utilised for the Le Fort cuts and then subsequently the

genioplasty.

Respiratory cysts are lined by a thin layer of ciliated pseudostratified columnar epithelium, normally found in sinuses or respiratory lining and was first described in the chin region following the use of the nasal hump and septal cartilage graft by Aufrucht in 1934 [2]. He also warned against leaving any attached mucosa on harvested bone or cartilage. Lesions are locally invasive with recurrence rate of up to 50% [4,5].

A number of differential diagnosis are possible for cystic lesions of the mandible and must be considered. These include dentigerous cysts, radicular cysts, ameloblastoma, odontogenic keratocysts, neoplastic processes as well as others and are listed in Table 1. A biopsy should be taken to establish a diagnosis. Differential diagnosis may also include odontogenic cysts containing pseudostratified ciliated columnar epithelium and are usually associated with mucous cells which are absent in these specimens [3].

We speculate that sinus mucosa was transported via the saw blade used to make the Le Fort cuts although this cannot be confirmed. Clinically the cysts may present with numerous symptoms, including pain, discharge following infection, bony expansion, or may simply be an incidental finding during investigation of other pathologies.

Radiographically, the cyst may present as a unilocular or multiloculated cyst with bony expansion. Initial investigations include an OPT and subsequently a CT of the mandible may be required to fully assess the extent of the cyst and local destruction.

The cysts are locally invasive and the treatment of choice is enucleation, as with any cystic lesion. However, marsupialization may be preferred due to the size of the lesion as demonstrated in Case 1. Incomplete removal of the cyst lining may result in recurrence with a recurrence rate of up to 50% [6-8].

With regards to the cases described above and the time period since the initial osteotomies, it was impossible to say whether or not bone graft from the maxilla was utilized for the genioplasty. We assume the same saw blade was used for both the Le Fort osteotomy and the genioplasty, allowing for iatrogenic implantation of sinus lining in the mandible.

Conclusion

Mandibular cysts are a relatively common occurrence. However in patients presenting following previous orthognathic surgery, the differential diagnosis should include the rarer respiratory cyst. Treatment of this benign pathology is complete enucleation and recurrence should be considered with incomplete enucleation cases.

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