Respiratory implantation cyst of the mandible following orthognathic surgery

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Abstract

A cyst following implantation of respiratory epithelium during surgical procedures has been documented only rarely. A variety of names have been attached to this lesion: respiratory mucocele, respiratory implantation cyst, surgical ciliated cyst, and surgical (implantation) cyst. In seven prior case reports, the interval between the initiating surgical procedure and diagnostic biopsy of the resultant well-circumscribed radiolucency and histopathological demonstration of distinctive pseudostratified ciliated columnar epithelial lining has varied from 4 to 40 years. In the case reported here, chin augmentation used "residual maxillary bone" as the donor tissue and likely transplanted sinonasal mucosa into the mandible during orthognathic surgery, resulting in a painful anterior mandibular cyst lined by respiratory epithelium that was ablated 16 years later.

Keywords: Mandibular cyst, respiratory mucocele, respiratory implantation cyst, surgical ciliated cyst

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Introduction

The eventuation of a cyst following implantation of respiratory epithelium during surgical procedures is a rare event. Literature review located only seven cases of ciliated implantation cysts occurring in the mandible. Decades may pass before sufficient enlargement or secondary infection calls such a radiolucent lesion to clinical attention. In the case reported here, chin augmentation used "residual maxillary bone" as the donor tissue and likely transplanted sinonasal mucosa into the mandible during orthognathic surgery, resulting in a painful anterior mandibular cyst lined by respiratory epithelium that was ablated 16 years later.

Case Report

In 1992, a 14-year-old white male underwent surgical correction for developmental, musculoskeletal, dentofacial deformity. The preoperative diagnosis included vertical maxillary excess with lip incompetence: excessive show of maxillary teeth at rest and animation, "gummy smile," revealing approximately 10mm of anterior teeth at rest and resultant gingival inflammation, Class II mandibular retrognathia with significant microgenia, masticatory dysfunction, and four bony impacted third molars. Cephalometric radiological evaluation and orthodontic study model analysis confirmed the clinical diagnoses. Following presurgical orthodontic treatment the patient underwent surgical correction consisting of a LeFort I maxillary osteotomy, with 8mm of vertical ostectomy, horizontal osteotomy of the mandible with advancement of the inferior border and removal of four bony impacted third molars. The osteotomized segment of the mandible was advanced anteriorly by 7mm and stabilized. The bony ledge of the osteotomized advanced segment was grafted, with bone removed from the ostectomy of the maxilla. There were no immediate surgical complications.

A few years prior to 2008, progressive pain led to detection of an anterior "mandibular cyst" that was biopsied at a facility not recalled by the patient and so could not be pursued. After biopsy, no further treatment was sought because his pain was relieved. The patient presented in early 2008 with acute swelling, pain, and purulent discharge, from what appeared to be an anterior mandibular vestibular abscess. On further clinical examination, the teeth were found to be stable, although there were cervical restorations on the incisor teeth and poor oral hygiene due to pain. Endodontic pulp testing found some of the teeth to be nonvital. Radiological studies revealed a large anterior mandibular cyst apical to the mandibular incisors extending from the right mandibular canine to the left mandibular canine (Figure 1a). The apices of the mandibular teeth were involved in the superior extent of the radiolucent area (Figure 1b). Computerized tomography placed the lesion in the marrow space of the bone (Figure 1c,d) with cortical perforations (Figure 2). An incisional biopsy was performed along with incision and drainage of infection. Following the biopsy and eradication of infection, in view of the patient's military obligations and anticipated impediments to follow-up, prophylactic endodontic therapy was performed on all six mandibular teeth, prior to complete enucleation and peripheral
Histological examination of the biopsy (and later a complete curettage specimen) revealed granulocytes, lymphocytes, and plasma cells scattered in an edematous stroma that supported a pseudostratified ciliated columnar respiratory-type epithelium (Figure 3). Unremodeled chondro-osseous remnants of the graft, as found by Lazar, \cite{1} were not observed.

At the time of definitive curettage, 49 days after biopsy, the cyst was confirmed to have perforated both the buccal and lingual plates of the anterior mandible. He was grafted with rH-bmp (recombinant human bone morphogenic protein, Medtronic®) at the time of treatment and was without dehiscence or symptoms at the six-month follow-up.

**Discussion**

The majority of reported cases of surgical ciliated (implantation) cysts have been in the maxilla following radical sinus surgery in people of Asian ancestry, where the incidence has been reported to be up to 20%. Only a few of these have been secondary to orthognathic surgery. The presumed mechanism for these cysts forming after orthognathic surgery is that the sinus and/or nasal mucosa is trapped in the wound during closure. Enhanced by the highly vascularized environment of the recipient site created during the healing process, the differentiated sinonasal epithelium trapped in the wound proliferates to create the ciliated implantation cyst. \cite{2,3} A variety of names have been attached to this lesion: respiratory mucocele, \cite{3} surgical ciliated cyst, \cite{4} surgical (implantation) cyst, \cite{5} and respiratory implantation cyst. \cite{1}

While history of the original procedure was not known at the time of pathological analysis, the clue to the diagnosis in this case was the 2008 panoramic radiograph (Figure 1a). It indicates prior orthognathic surgery, a LeFort I, due to the presence of osteotomy-associated metallic plates at the nasal aperture, and chin surgery with stabilizing ligature wires below the cuspid region. From an initial literature review, we postulated that chin augmentation using sinonasal bone and/or cartilage as the donor tissue likely transplanted the mucosa into the mandible during orthognathic surgery. \cite{1,2,3,4,5,6} Subsequently, records of the original surgery in 1992, supplied by one of the authors who was at the procedure, (BNE), confirmed that hypothesis.

The development of respiratory implantation cysts lined by a thin layer of ciliated, pseudostratified columnar epithelium in the chin region have been described following use of the nasal hump and septal cartilage as described by Aufricht, in 1934. \cite{6} Aufricht himself warned against leaving any attached mucosa on bone or cartilage harvested from the nose. Others agree that this unusual complication can be avoided by removal of all adherent nasal mucosa from the graft. \cite{1,3}

Our review located only seven cases of ciliated implantation cysts in the mandible (Table 1). These cases were all reported secondary to chin augmentation following genioplasty.

procedures, utilizing osteocartilaginous nasal grafts, that is, simultaneous LeFort I, II or III osteotomy and genioplasty. It is proposed that transplanted respiratory epithelium, attached to the sinonasal-derived graft material, proliferates in the favorable healing environment of the grafted site, essentially creating a sinus cavity in the marrow space. In only one of the seven reported cases, the cyst was located anterior to the bone, leaving the underlying osseous structure intact. Decades may pass before the cyst is detected, as illustrated by cases C and G in (Table 1). Their clinical presentation ranges from asymptomatic to an indolent enlargement until it becomes an "aesthetic hindrance" or acute swelling for several days, due to secondary infection. Their common histopathological feature is the distinctive ciliated respiratory-type mucosa. Postoperative resolution varies from weeks to years.

No bone graft was installed during the gliding genioplasty done years before the small cystic mandibular lesions reported by Bourgeois and Nelson. The authors postulate sinus mucosa was transplanted on the saw blade following the LeFort I osteotomy and recommend "meticulous cleaning of the saw blade" or using a second blade.

A likely eighth case is that of Caputy and Flowers, who describe a patient "with migration of the chin implant" following an autologous nasal cartilage graft employed in a chin augmentation 30 years previously. On dissection down to the implant, they encountered "an abscess pocket containing approximately 20ml of thick, white, totally homogeneous, nonodiferous pus." They removed the cartilaginous implant and curetted the area, which they thought to be a "sterile abscess," although subsequent cultures were positive for *Staphylococcus mitis*. Unfortunately, no specimen was sent for histopathology.

Pseudostratified ciliated columnar epithelium is normally found lining the sinuses and respiratory tract and is therefore remarkable in a mandibular cyst. Important to the differential diagnosis in the present case, maxillary and mandibular odontogenic cysts containing pseudostratified, ciliated, columnar epithelium have occasionally been reported in epithelial linings of odontogenic inflammatory cysts (radicular cysts) and developmental cysts (dentigerous and primordial cysts) of the maxilla and mandible. Such ciliated cells are assumed to arise through metaplasia or prosoplasia and are always accompanied by mucous cells, not seen in this patient's specimens. Further against the endodontic origin of the cyst are the large size of the lesion, significant infection, and some teeth remaining viable although encroached upon by the lesion.

As a further pathology cautionary note, such a case could easily be misdiagnosed as inflamed maxillary sinus membrane if the exact location of the cyst is not specified in the history submitted with the specimen.
Table/Figures:

<table>
<thead>
<tr>
<th>Case</th>
<th>Author(s) &amp; Report year</th>
<th>Patient age at detection</th>
<th>Symptoms and findings</th>
<th>Size</th>
<th>Radiology</th>
<th>Initial procedure type</th>
<th>Years since previous procedure</th>
<th>Operative findings: Content type of cyst lining</th>
<th>Followup</th>
</tr>
</thead>
<tbody>
<tr>
<td>A</td>
<td>Lazar 2005</td>
<td>24/Male</td>
<td>Red swelling of submental region, local infection with abscess formation</td>
<td>4.5 x 1.5 cm</td>
<td>Ovoid apical lucency with well-defined border</td>
<td>Rhinoplasty and osteoplasty using nasal hump</td>
<td>5</td>
<td>Respiratory epithelium remnants of transplanted cartilaginous graft</td>
<td>Long-term external drainage, osteocartilaginous, okay chin contour after 2.5 years</td>
</tr>
<tr>
<td>B</td>
<td>Anastasov and Lee 1999</td>
<td>53/Male</td>
<td>Gradual enlargement over 6 months. Became an “esthetic hindrance”</td>
<td>5 x 6 cm</td>
<td>Anterior mandible oval lucency with sclerotic border affecting anterior cortex and mandibular bone</td>
<td>Reduction rhinoplasty, septoplasty, and chin augmentation</td>
<td>39</td>
<td>Not specified</td>
<td>“Suction” fluid, “Soft like” cavity covered by a thin ciliated pseudostratified columnar epithelium</td>
</tr>
<tr>
<td>C</td>
<td>Imholte and Schwartz 2001</td>
<td>50/Male</td>
<td>Acute swelling of lower lip and lower lateral vestibule for several days</td>
<td>Not specified</td>
<td>Unilocular lucency in synovial cyst between roots of canines</td>
<td>Septorhinoplasty after an accident with simultaneous chin augmentation using nasal bone and cartilage</td>
<td>4</td>
<td>Drained pus, primarily of soft tissues anterior to the mandibular symphysis. Creating a depression in anterior cortex. Fibrous connective tissue cyst wall. Lined by pseudostratified ciliated columnar epithelium</td>
<td>“Uncertain recovery”</td>
</tr>
<tr>
<td>D</td>
<td>Koutlas 2002</td>
<td>34/Female</td>
<td>Swelling, mild discomfort, unilateral draining fistula</td>
<td>Not specified</td>
<td>Radiodense multilocular cystic lesion</td>
<td>Orthognathic surgery of maxilla and mandible</td>
<td>23</td>
<td>Cysts lesion lined by respiratory epithelium, notable numbers of eosinophils in the cyst wall. Milky white semi- transparent aspirate, perforation of lingual cortex. Only the lesion between 21 and 22 had respiratory-type ciliated epithelium</td>
<td>Continues irrigation from sinus for 8 months</td>
</tr>
<tr>
<td>E</td>
<td>Bourgey et al. 2005</td>
<td>24/Female</td>
<td>Two asymptomatic radiolucent areas, unchanging over 32 months</td>
<td>2 mm area 30 &amp; 25 10 mm near teeth 21 &amp; 22</td>
<td>Two radiolucencies on a panoramic film taken for unrelated trauma, independent of space of teeth</td>
<td>Multiple maxilloturbinotomy and gingly, genoplasty without graft</td>
<td>4</td>
<td>Not specified</td>
<td>Not specified</td>
</tr>
<tr>
<td>F</td>
<td>Nafti and Hooker 1984</td>
<td>55/Female</td>
<td>3 years’ ulceration and episodic pain in the mandibular sinus, 18 months gradual increase in swelling</td>
<td>7.0 x 3.5 cm</td>
<td>Smooth-bordered lytic defect of mandible latal plate involving most of the alveolar and basal bone and displacing the root of the left mandibular canine</td>
<td>Reduction rhinoplasty with chin augmentation with subperiosteal enucleation of maxilla bone and cartilage harvested from the nose</td>
<td>15</td>
<td>Pseudostratified ciliated columnar epithelium. “For the most part”</td>
<td>Not specified</td>
</tr>
<tr>
<td>G</td>
<td>Kelly et al. 2000</td>
<td>56/Female</td>
<td>Painful swollen chin</td>
<td>2.2 cm greatest dimension</td>
<td>Underlying osseous structure intact Cyst uniformly radiolucent</td>
<td>Simultaneous rhinoplasty and genioplasty</td>
<td>40</td>
<td>Opaque viscous fluid, pseudostratified columnar epithelium</td>
<td>Completely healed after 4 weeks</td>
</tr>
</tbody>
</table>

Table 1. Summary of the seven cases of ciliated implantation cysts in the mandible reported secondary to chin augmentation following genioplasty procedures utilizing osteocartilaginous nasal grafts.

Figure 1: Panoramic (a) and spot film (b) views depict a wellcircumscribed, lucent anterior mandibular lesion with lobulated sclerotic border. Anteroposterior (c) and horizontal (d) CT cuts placed the lucency in the marrow space, with perforation of anterior cortex.
Figure 2: A 3-D reconstruction displays the cortical perforation in the anterior mandible.

Figure 3: Ciliated pseudostratified columnar epithelium of a respiratory type exclusively constituted the lining of the cyst.

References