Maxillary keratocystic odontogenic tumor with sinus involvement: a multidisciplinary approach

Drs. Brian Trava, Neil Thoman, Sharon Brooks, Harry Katz, Mark Persky, and Kathleen Nagy treat an aggressive tumor with comprehensive communication between different modalities of medicine and dentistry

Introduction

The following is a case report of a long-standing lesion diagnosed using 3D CBCT imaging. The ease of cone beam accessibility allows endodontic specialists to diagnose, collaborate, and deliver prompt multidisciplinary treatment to our patients. In 2005, the World Health Organization (WHO) reclassified the odontogenic keratocyst (OKC) into the tumor category as a keratocystic odontogenic tumor (KCOT). The lesion is characterized as being aggressive with a high reoccurrence rate.

History

A 60-year-old Caucasian female was referred from her general dentist with a chief complaint of diffuse pain in the maxillary right side of her face. There was some discomfort upon chewing. The patient reported having pain and a root canal completed on tooth No. 3 in 1994. The symptoms continued, and the patient sequentially had an apicoectomy, extraction, and a fixed bridge spanning from teeth Nos. 2-4. The patient also remembered having some type of hard mass removed from her sinus in the 1980s. The patient reported a history of having a bad gag reflex, making it difficult to take dental radiographs throughout her lifetime.

Oral exam

Teeth Nos. 2-6 responded to a cold test using an ice chip. Tooth No. 2 was tender to percussion. All mandibular teeth tested within normal limits (WNL). Gingival tissue looked WNL. The right tuberosity area was depressible.

Radiographic exam

A traditional periapical film was attempted and failed due to the patient's gag reflex. A Planmeca 2D panorex was taken. The panorex demonstrated the maxillary right tuberosity was radiolucent and expanded, plus there was degenerative joint disease in the right TMJ (Figure 1). A Planmeca CBCT 8 x 8 image at 90kV/12mA/12.3s was rendered (Figure 2). The CBCT DICOM images were uploaded to BeamReaders for radiographic interpretation. There was a gross expansion

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Harry Katz, MD, devotes his practice to the general care of the ears, nose, and throat with particular emphasis on diagnosis, treatment, and surgery for nasal and sinus diseases. He is a graduate of Columbia University and then went on to complete his medical school, internship, and residency training at New York University Hospital and Medical Center in New York. He maintains a practice in Midland Park, New Jersey.

Sharon Brooks, DDS, earned her dental degree from the University of Michigan in 1973, along with MS degrees in Oral Diagnosis and Radiology (1976) and Radiological Health (1984). Dr. Brooks is a Diplomate of the American Board of Oral and Maxillofacial Radiology and served 5 years as a director of the Board and past president. Dr. Brooks has been working with CBCT since 2004. She joined BeamReaders in 2010.

Mark Persky, MD, is an ENT-otolaryngologist in New York, New York and is affiliated with multiple hospitals in the area, including Mount Sinai Beth Israel and Mount Sinai St. Luke's-Roosevelt. He received his medical degree from State University of New York Upstate Medical University and has been in practice for 42 years.

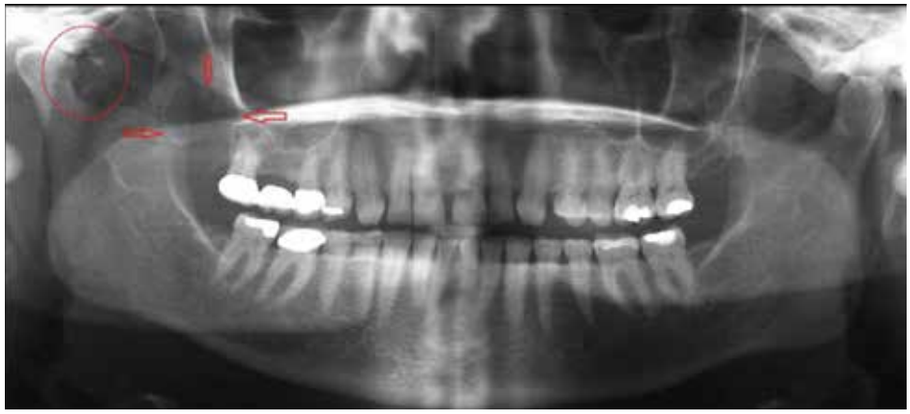

Figure 1: Traditional panoramic view comparing left and right sides

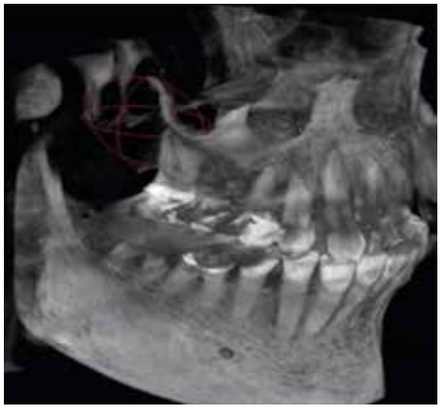

Figure 2: Planmeca 3D image with volume of area quantified

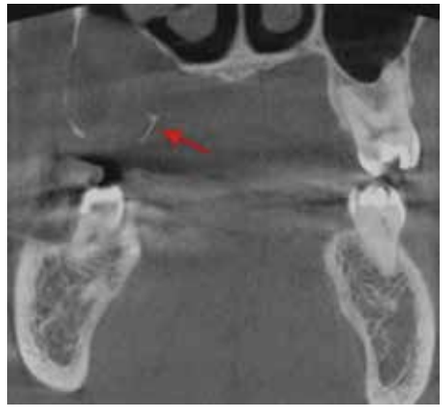

Figure 3: Frontal view shows thin cortex and expansion

of the right maxillary tuberosity distal to tooth No. 2 with extreme thinning of the cortex on all sides (Figure 3). A radiolucent area extended medially into the palate and posteriorly into the pterygomaxillary area (Figure 4). Locules extended anteriorly to the second molar. The floor of the sinus was elevated by the lesion distal to tooth No. 2 in its normal location anterior to the tuberosity. The medial wall of the right sinus was missing, although this may have been a result of the previous surgery. The margins of the lesion appeared curved and well-defined, with an occasional suggestion of fine wispy septa within the lesion (Figure 5).

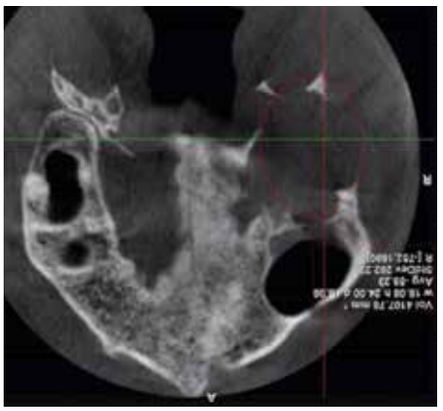

Figure 4: Axial section shows expansion into the palate ant pterygomaxillary area

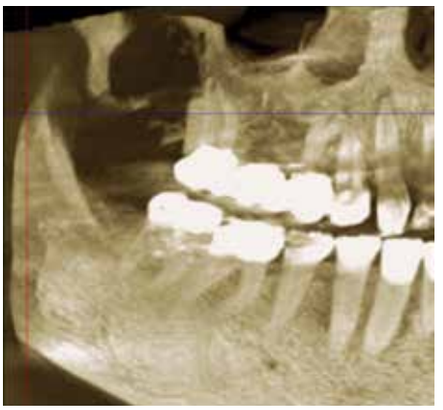

Figure 5: Planmeca 3D enhancement can provide more diagnostic detail

Differential diagnosis

The position of the lesion in the right maxillary tuberosity rather than the maxillary sinus led us to believe it was from a history of odontogenic origin. The lesion gave the appearance of a keratocystic odontogenic tumor. Since the lesion was located only to the maxillary jaw, ameloblastoma would be more likely than myxoma, central giant cell lesions, and nevoid basal carcinoma.

The patient was referred to the oral surgeon for consultation and treatment. A preliminary distal wedge excisional biopsy was performed and sent for biopsy.

The biopsy showed stratified squamous epithelium covering a core of well-vascularized fibrous connective tissue with a dense infiltrate of neutrophils, lymphocytes, and plasma cells. The pathologist's impression was acute inflammatory reaction. The patient was referred to an ENT for consultation and treatment. The patient underwent a right Caldwell-Luc procedure. A cystic mass with inspissated secretions was excised. Fragments of benign, partially squamous, and sinonasal type epithelium-lined cyst wall with dense fibrosis marked inflammation.

After discussion, with correlation to radiographic results, the final diagnosis was keratocystic odontogenic tumor (KCOT/OKC).

Discussion

The World Health Organization reclassified the odontogenic keratocyst as a keratocystic odontogenic tumor in 2005. Clinically, this presents as a swelling with or without pain, less frequent in the maxilla.[1] The maxillary involvement ranges from 16.4% to 23.5%, and 1% occurs in the maxilla with sinus involvement.[2] The lesion typically grows in an anterior to posterior region.[3] The patient presented a history of both a failed root canal and apicoectomy and history of previous sinus surgery, accompanied with the sacristy of radiographic history due to difficulty with gagging. There may be a remote possibility that the present lesion may have been recurrent, characteristic with the KCOT, or had started a slow progression after the original sinus surgery in 1980 in which a hard mass was removed. This could have been an associated tooth, which has been reported. The tumor has been reported to have an equal distribution of occurrences from decades 3 to 7.5 The lesion has a reoccurrence rate of 29% to 58%.[1,6]

The lesion is cystic in nature with a thin lining, described as neoplastic in nature with orthokeratinized and parakeratinized variants, and is closely associated with the ameloblastoma.[7,8]

Having a multidisciplinary approach to diagnosing and treating patients provides patients such as this with the most efficient treatment and predictable prognosis. Patients that have special needs, such as gag reflexes, autism, and Down's syndrome, cannot tolerate intraoral radiographs. From an endodontic standpoint, many patients are referred for diagnosis due to pain. An 8 x 8 cm CBCT that can render both a traditional 2D and 3D image offers the advantage of bilateral orthognathic and sinus diagnosis capability. Being able to select an area on a 2D panorex allows the clinician to scout a particular quadrant, or from TMJ to TMJ. Radiographic interpretation for diagnosis will always have its academic foundation based on traditional radiology. Planmeca 3D ProMax® technology provided expediency to upload radiographic files to the cloud or via the Internet for immediate diagnosis and treatment from the appropriate specialists. This allowed much clearer communication between the endodontic specialist, the maxillofacial surgeon, and the ENT.

Conclusion

The KCOT is an aggressive tumor with a high reoccurrence rate. Diagnosis requires comprehensive communication from different modalities of medicine and dentistry. CBCT has definitively enhanced these avenues for a more thorough treatment approach for patients. EP

Acknowledgements

Alexander Filatov, MD, is a pathologist, has 16 years of experience and practices in Anatomic Pathology & Clinical Pathology and Anatomic Pathology.

Dr. Stanley Kerpel, DDS, specializes in dentistry, oral and maxillofacial pathology, and pathology, and currently treats patients in Flushing, New York and Bedford, New York.

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