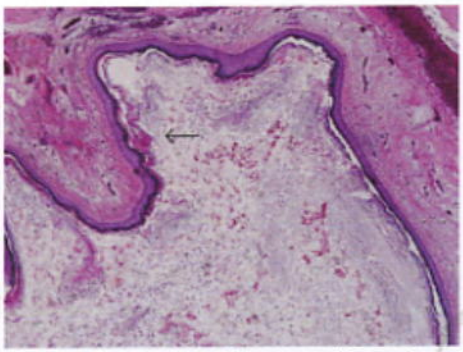

FIGURE 3. Histologic finding (hematoxylin-eosin stain, original magnification ×40). Cyst filled with keratotic material including keratin pearl (arrow).

differential diagnostic lesions that develop in the facial area include dermoid cyst, lipoma, hemangioma, neurofibroma, and others. Each case can be easily differentiated by radiologic and histologic examination.[1]

In case of a large epidermal cyst, various examinations such a ultrasonography, CT, magnetic resonance imaging (MRI), and other radiologic examinations can be performed to differentiate it from other tumors.[4] Ultrasonography is considered a cost-effective and easily used diagnostic tool compared with MRI. On MRI, epidermal inclusion cyst shows low signal intensity on T1-weighted images and high signal intensity on T2-weighted images. Also, absence of contrast enhancement within tumors on T1-contrast enhancement images can be observed.[4]

Of all the methods described for treating epidermal inclusion cysts, such as treatment with Solcoderm (a copper ion and acid solution), minimal excision, punch incision, and, recently, extraction and curettage using an endoscope, traditional total excision is generally preferred.[9–12] It is the method of choice and the surest way to prevent recurrence.[10,13] It thus saves the patient further operations, time, and money.[14]

During the excision, intraepidermal layer should be included for complete excision and to prevent the recurrence. Malignant change of benign epidermoid cysts is rare, but their prognosis remains poor.[15] Therefore, all excised epidermal cysts should undergo a pathologic examination to exclude malignancy.[16]

Epidermal inclusion cysts of the face are common and can be easily encountered in clinical practice. They are usually small, with sizes varying from a few millimeters to a few centimeters. Few giant epidermal cysts have been reported so far, and giant epidermal cyst of the face has never been reported. We have experienced a rare case of giant epidermal inclusion cyst of the face and report this case with a brief review of literature.

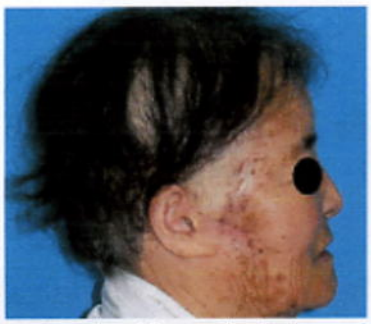

FIGURE 4. Postoperative 10-month photograph shows good cosmetic result without recurrence.

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Peri-Implant Squamous Odontogenic Tumor

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Abstract: Squamous odontogenic tumor (SOT) is a benign, locally infiltrative intraosseous tumor composed of well-differentiated squamous epithelium in a fibrous stroma. It seems to derive from the epithelial rests of Malassez in the periodontal ligament space. It presents an odontogenic origin, involving both the upper and lower maxillary bone, mainly areas without teeth or connective tissue

of the odontogenic cysts. Clinically, SOT could be asymptomatic (3 cases), notwithstanding it is mainly characterized by pain, swelling, and tooth/teeth mobility. The most typical presentation of SOT is a slowly growing endobony lesion arising within a single periodontal location. Frequent misdiagnosis concerns either ameloblastoma and squamous cell carcinoma and fibroma. Since its first description in 1975, less than 50 cases have been identified. In light of the few reported cases, there are no consistently recorded clinical and radiographic features of SOT, and there is no predictable sex or site predilection. Diagnosis is predicated on recognition of the histopathologic features of SOT to obviate possible misdiagnosis of malignancy or ameloblastoma.

We report the first case of SOT that arose in the vicinity of an implant. Through a meticulous review of literature, we discuss current etiology, pathogenesis, and treatment.

Key Words: squamous odontogenic tumor, Malassez, conservative treatment, implant

Squamous odontogenic tumor (SOT) is a rare benign odontogenic neoplasm first described by Pullon and colleagues[1] in 1975. It is supposed to originate from the epithelial rests of Malassez that represent remnants of Hertwig epithelial root sheath, an epithelial membrane that guides root formation.[1–3] These epithelial rests are most numerous on the lateral surface root, although they may be found anywhere within the periodontal ligament.[4,5] Thus, typically SOT radiographic presentation is a triangular or semicircular radiolucent defect involving the lateral roots of erupted teeth.

Fewer than 50 cases had been reported in the literature.[1–57] We report the first case of a SOT that developed in a peri-implant location. Through a meticulous review of literature, we discuss the etiology, pathogenesis, and treatment of this rare disease.

CLINICAL REPORT

In February 2003, a 64-year-old man with a fracture of the tooth 45 and pain to percussion presented to our department. A fracture of the radicular part coupled with alveolar resorption has been diagnosed after a radiographic scan. The patient had previously submitted to avulsion of the aforementioned tooth. Three implants have been applied in zones 45, 46, and 47 as definitive prosthesis to achieve immediate rehabilitation. Although initially no radiolucent lesion has been noted, 2 months later he referred pain on the right jaw, and a radiotrasparent lesion developed next to 45 implant was identified after radiographic evaluation. One week later, spontaneous rejection of the implant occurred, and because pain and swelling extended to 44 and 43, we performed an endodontic therapy on 44 followed by an intralesional biopsy. Diagnosis of SOT has been made according to histopathologic examination. In December 2003, the patient underwent radical surgical excision and reconstruction by a mucoperiosteal buccal flap. The neurovascular pedicle and its canal were sacrificed because they were involved in the tumor. A final deep bone curettage followed by absorbable sutures has been performed (Fig. 1). During the 7 years' follow-up, the patient did not present recurrence both radiographically and clinically.

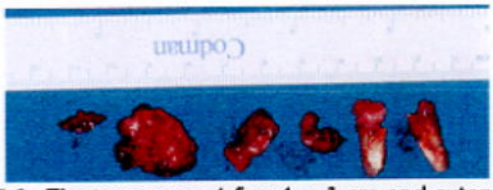

FIGURE 1. The tumor was 4.5 × 4 × 1 cm and extended from tooth 43 to 46. Photograph shows multiple fragments after radical bone curettage.

Histopathology

Five- to 6-μm-thick sections were prepared and stained with hematoxylin-eosin. Immunohistochemistry examination with the monoclonal antibody Ki-67 (clone MIB-1; Dako, Carpinteria, CA) revealed hyperkeratinized layers over the chorion and near the mature bony lamellar trabeculae. Jerkily small keratinized masses were visible in the cavity coated by a mature squamous epithelium. The squamous epithelium was organized in chords and islets into the chorion (Fig. 2 and 3). All these new formed epithelial structures were variable in dimensions, irregular or circle form, and without cytologic atypia. No typical or atypical mitotic aspect was observed. Necrosis was absent, whereas a moderate inflammation could be detected in the stromal background (Fig. 4). Epithelial residues of Malassez were identified positive by immunohistochemistry for cytokeratins AE1/AE3. Cellular proliferation index (MIB-1) was lower than 5% (Fig. 5).

DISCUSSION

Squamous odontogenic tumor is probably the rarest of all odontogenic tumors. According to the literature, only 41 prior cases of SOT were described and confirmed by clinical, radiographic, and

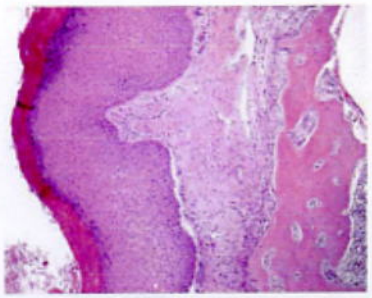

FIGURE 2. The epithelial squamous lamina near the trabecular bone is hyperkeratosis (hematoxylin-eosin, original magnification ×100).

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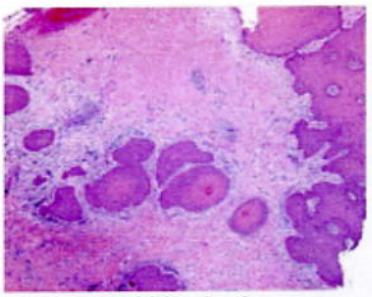

FIGURE 3. Multiple round islands of squamous epithelium into the chorion (hematoxylin-eosin, original magnification ×50).

histopathologic data. Although further 10 cases have been found, unfortunately their reports are not supported by necessary data, and a convincing diagnosis of SOT cannot be confirmed.[1–57] Analyzing the few published cases, it is still possible to draw some conclusions regarding the usual demographic, clinical, and radiographic presentation of the tumor (Table 1).[1–57]

The origin of SOT is still being debated. Although most of investigators believe that SOT arises from the epithelial rests of Malassez in the periodontal ligament space, someone proposed its origin from the rest of Serres or the overlying gingival epithelium.[1,5,11] The epithelial islands intimately associated with the apices of retained teeth suggest that the tumor could origin from the rests of Malassez.[7,11] On the other hand, someone proposed the rest of Serres as origin of the lesion after having observed proliferation of squamous epithelial islands in SOT.[10] However, there exist peripheral variants of SOT that are thought to arise from gingival surface epithelium or from remnants of the dental lamina (rests of Serres).[4,5]

From the literature review, it emerges that SOT affects those with ages ranging from 8 and 74 years (average, 39.4 years); males are little more affected than females, and white race is more frequently involved than is black race. Mandible involvement is similar to the maxilla (17 cases each); multiple locations occurred in 7 cases even if the primary site remains unknown. In particular, the most common locations for the development of SOT are the anterior maxilla (25%) and the posterior mandible (17%). It has not been possible to identify the exact location in 18% of the cases. When SOT presented in the mandible, it was most frequently associated with impacted third molars (9 cases),[12–14] whereas the remaining cases presented within the mandibular-first-second-molar region[14,15] and within the mandibular first premolar–canine region[48] with a relatively equal distribution of each. When maxilla was the primary site, the canine–first premolar area was the most frequently involved (Table 1).[14,16]

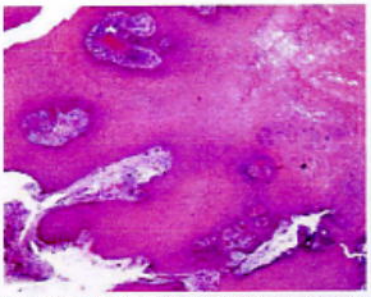

FIGURE 4. Wide foil of SOT devoid of cytologic atypia and mitosis (hematoxylin-eosin, original magnification ×50).

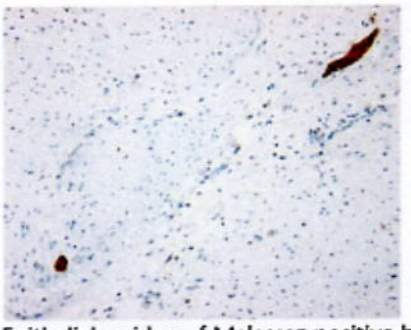

FIGURE 5. Epithelial residue of Malassez positive by immunohistochemistry for cytokeratins AE1/AE3 (original magnification ×200).

Clinically, SOT can be asymptomatic, notwithstanding it is mainly characterized by pain, swelling, and tooth/teeth mobility. The most typical presentation of SOT is a slowly growing endobony lesion arising within a single periodontal location. Mobility of the peritumoral teeth is the predominant sign. Symptoms vary, but they may include moderate pain and swelling of the surrounding soft tissue. Interestingly, it has been observed that SOT behaves more aggressively when found in the maxilla than in the jaw,[17,19] probably owing to the anatomy of the region, and the porous, medullary nature of bone.[16] Although histologically benign, the tumor presents a locally destructive behavior; thus, it could be explained why root resorption has been found only in 1 case (Table 2).[17,19,27]

The lesion may present as an incidental findings on routine dental radiographs. However, the radiotrasparent appearance was the most frequent radiologic presentation even if radiographic findings are far from being pathognomonic. The radiolucent areas are not well defined and consist of triangular or vertical periodontal bone loss.

In light of the few reported cases, there are no consistently recorded clinical and radiographic features of SOT, and there is no predictable sex or site predilection. Diagnosis is predicated on recognition of the histopathologic features of SOT to obviate a possible misdiagnosis of malignancy or ameloblastoma.[14] Diagnostic errors could result in overtreatment and disfiguring surgery.

Histologically, islands of benign squamous epithelium surrounded by mature connective tissue stroma without evidence of peripheral columnar cells, palisading nuclei, or stellate reticulum represent the main features of SOT.[7] Its epithelial islands are very similar to squamous metaplasia observed in ameloblastomas; hence, the absence of peripheral columnar cells and palisading nuclei allows differentiating these 2 neoplasms.[7] Diagnosis of carcinoma (mainly squamous cell carcinoma) can be excluded as there is no malignant cell present.

Other features such as keratin pearls, microcysts, and intraepithelial calcifications may often be observed in SOTs, as well as some of the epithelial islands with circular areas of increased fibroblastic activity and fibrous hyalinization, reason for which remains still unknown. This last aspect may suggest a connective tissue reaction to the epithelial proliferation,[7] even though it can also be inferred that the unusually large hyalinized juxtaepithelial areas could be an important inherent aspect of the neoplastic process of SOT.[13]

The benign nature of the SOT suggests a conservative surgical treatment with bone curettage that resulted in few recurrences.[3,17,20] Further local enucleation and scaling of adjacent teeth have been used successfully.[1,16] Recurrences appear to respond to secondary conservative procedure such as local curettage and extraction of the involved teeth.[16,18,21] Very rarely, aggressive surgical

TABLE 1. Distribution of the Identified Squamous Odontogenic Tumors According to Age, Sex, Race, and Location

Clinical Report	Age, y	Sex	Race	Location
1	23	F	Black	Multiple, both the maxillas
2	11	M	White	Anterior maxilla
3	19	M	White	Posterior mandible
4	31	F	White	Anterior maxilla
5	42	F	White	Anterior maxilla
6	29	M	White	Posterior mandible
7	26	M	Black	Anterior maxilla
8	65	M	Unidentified	Anterior maxilla
9	26	F	Black	Multiple, 4 quadrants involved
10	22	F	Black	Both maxilla and mandible
11	59	M	White	Anterior mandible
12	66	F	White	Superior maxilla
13	59	M	White	Anterior mandible
14	60	F	Unknown	Posterior mandible
15	29	M	Unknown	Posterior mandible
16	30	F	White	Posterior maxilla
17	26	F	White	Posterior mandible
18	67	F	White	Unspecified
19	24	F	White	Anterior mandible
20	26	M	Black	Multiple bilateral location in maxilla, carcinoma of mandible
21	61	M	Unknown	Maxilla
22	51	F	Unknown	Mandible
23	19	M	Unknown	Mandible
24	26	M	Black	Multiple, both maxilla and mandible
25	41	M	Unknown	Anterior mandible
26	56	M	Unknown	Maxilla
27	29	M	Unknown	Multiple
28	25	M	Unknown	Multiple
29	38	M	Unknown	Anterior maxilla
30	56	F	Unknown	Mandible
31	46	M	Unknown	Upper right maxilla
32	39	M	Unknown	Posterior mandible
33	74	M	Unknown	Maxilla
34	32	M	Unknown	Anterior maxilla
35	56	M	Unknown	Posterior maxilla
36	57	M	Unknown	Anterior mandible
37	25	M	Unknown	Mandible
38	42	F	Unknown	Upper maxilla
39	45	M	Black	Maxilla
40	25	Unknown	Unknown	Mandible
41	32	M	White	Mandible

treatment has been used: hemimandibulectomy (3 cases),[18,19,22] partial maxillectomy (1 case),[23] and total maxillectomy (1 case).[24] Although Ide and colleagues[25] noted that SOT could transform into a malignant neoplasm such as squamous cell carcinoma, actually from the global literature review the conservative surgical approach seems to be the mainstay of treatment. Maxillary lesion may necessitate slightly more extensive procedure to ensure resolution.[26]

The case we presented is peculiar because it was the first case of SOT that developed in a peri-implant location. The reason behind the wide surgical demolition, sacrificing the neurovascular pedicle and its canal, was due to their involvement into the tumoral mass because a delay in diagnosis had allowed the tumor to grow undisturbed. Thus, the physician needs to watch out for clinical signs of SOT and to be aware that this kind of tumor could be found in a peri-implant setting too, as our report showed.

TABLE 2. Clinic, Treatment, and Follow-Up of the 41 Squamous Odontogenic Tumors Currently Described in the Medical Literature

Clinical Report	Clinic	X-Ray Findings/Features	Follow-Up	Treatment
1	Teeth mobility	Radiotrasparent lesion	No recurrence after 5 y	Odontectomy and alveoloplasty
2	Swelling	Radiotrasparent lesion	No recurrence after 5 y	Surgical excision, tooth extraction
3	Asymptomatic	Radiotrasparent lesion	No recurrence after 11 y	Surgical excision
4	Teeth mobility	Loss of trabecular bone	No recurrence after 1 y	Surgical excision
5	Teeth mobility	Loss of alveolar bone	No recurrence after 5 y	Surgical excision
6	Bone loss	Radiotrasparent lesion	No recurrence after 18 y	Surgical excision
7	Pain and tooth mobility	Radiotrasparent lesion	Unknown	Unknown
8	Teeth mobility	Multilocular area	No recurrence after 7 mo	Partial maxillectomy
10	Teeth mobility	Radiotrasparent lesion	No recurrence after 3 mo	Surgical excision, hemimandibulectomy
11	Swelling, teeth mobility	Unknown	No recurrence after 1 y	Surgical excision
12	Swelling, teeth mobility	Triangular radiotrasparent lesion	No recurrence after 2 y	Surgical excision, curettage
13	Asymptomatic	Radiotrasparent lesion	Unknown	Surgical excision
14	Pain	Radiotrasparent lesion	Unknown	Surgical excision
15	Discomfort	Radiotrasparent lesion	Unknown	Surgical excision
16	Asymptomatic	Radiotrasparent lesion	Unknown	Surgical excision, tooth extraction, curettage
17	Unknown	Triangular radiotrasparent lesion	Unknown	Surgical excision
18	Increased thermic sensibility	Radiotrasparent lesion	No recurrence after 5 y	Surgical excision
19	Unknown	Unknown	No recurrence after 8 mo	Surgical excision
20	Asymptomatic	Radiotrasparent lesion	Unknown	Surgical excision
21	Unknown	Radiotrasparent lesion	Unknown	Surgical excision
22	Unknown	Radiotrasparent lesion	Unknown	Surgical excision
23	Unknown	Triangular radiotrasparent lesion	No recurrence after 2 y	Hemimandibulectomy
24	Asymptomatic	Radiotrasparent lesion	No recurrence after 2 y	Hemimandibulectomy
25	Unknown	Radiotrasparent lesion	Unknown	Surgical excision
26	Unknown	Irregular radiotrasparent lesion	Unknown	Partial surgical excision
27	Unknown	Bone loss	Unknown	Curettage
28	Unknown	Triangular radiotrasparent lesion	Unknown	Radical alveolectomy
29	Unknown	Triangular radiotrasparent lesion	Unknown	Surgical excision
30	Increased thermic sensibility	Radiotrasparent multilocular lesion	Unknown	Surgical excision
31	Unknown	Radiotrasparent lesion	No recurrence after 7 y	Surgical excision
32	Unknown	Radiotrasparent multilocular lesion	No recurrence after 1 y	Surgical excision
33	Unknown	Negative	No recurrence after 3 mo	Surgical excision
34	Unknown	Unknown	Unknown	Surgical excision
35	Unknown	Radiotrasparent unilocular lesion	No recurrence after 5 y	Surgical excision
36	Teeth mobility	Unknown	No recurrence after 18 mo	Unknown
37	Unknown	Radiotrasparent unilocular lesion	Unknown	Unknown
38	Unknown	Unilocular radiotrasparent lesion	Unknown	Unknown
39	Unknown	Radiotrasparent lesion	No occurrence after 18 mo	Surgical excision
40	Positive pulpar test vitality	Unilocular radiotrasparent lesion	No recurrence after 6 y	Surgical excision
41	Pain and swelling	Radiotrasparent lesion	No recurrence after 3 y	Surgical excision and curettage

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Nonossifying Fibroma Secondary to Aneurysmal Bone Cyst in the Mandibular Condyle

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Abstract: Nonossifying fibromas (NOFs) are benign lesions that unusually occur in the mandible. Nonossifying fibromas are asymptomatic and spontaneous resolution at skeletal maturity. Nonossifying fibromas associated with aneurysmal bone cyst (ABC) are very rare. In this clinical report, NOF secondary to ABC in the mandibular condyle was reported; however, it presents different clinical behavior than the usual NOF. In this case, severe destruction in the mandibular condyle as a characteristic of NOF was seen. In the follow-up period, no recurrence was seen subsequent to treatment of lesion with complete resection. Treatment of NOFs with secondary ABC would require aggressive intervention than the treatment of usual NOF.

Key Words: Nonossifying fibroma, mandible, aneurysmal bone cyst, complete excision

Nonossifying fibromas (NOFs) are benign lesion that belongs to a subgroup of fibro-osseous lesions. Nonossifying fibroma lesions occur most often in the distal femur, proximal and distal tibia, and fibula, but unusual in mandible. Nonossifying fibroma is a benign, well-marginated lesion of the bone composed of spindle-shaped cells with interspersed multinucleated giant cells and foamy histiocytes, often arranged in a storiform pattern. Radiographic evaluation showed that NOFs occurred in 30% of skeletally immature individuals. Radiographically, NOFs and fibrous, cortical defects appear as eccentric, well-defined, radiolucent, circular or oval lesions with smooth lobulated edges and surrounding sclerotic bone.[1] Nonossifying fibromas are asymptomatic unless they have reached a large size and are unrelated to trauma history.[2] Because most lesions are asymptomatic and there is spontaneous resolution at skeletal maturity, they do not require surgical intervention. These lesions may occur because of a disturbance of growth or an aberrance of calcification compared with a true neoplasm. The diagnosis of benign fibro-osseous lesions and further differentiation of the lesions within this group are based on clinical, radiographic, and histopathologic views. Aneurysmal bone cyst (ABC) is a pseudocyst found in blood-filled spaces in a connective tissue stoma containing multinucleated giant cells. The etiology and pathogenesis of ABC remain unknown, although the lesion is generally regarded as reactive.[3] One of different theories considers that ABCs are secondary lesions related to degeneration of a preexisting bone lesion such as the central giant cell granuloma (CGCG), fibrous dysplasia, or ossifying and cementifying fibromas.[4] Aneurysmal bone cysts more commonly occur in long-bone metaphysis and vertebra.[5] Within the craniofacial complex, ABCs are most common in the mandible, followed by the maxilla.[3] The body and the mandibular ramus are the main locations, with rare clinical reports in the coronoid process and the mandibular condyle.[6]

CLINICAL REPORT

A 17-year-old girl was referred to our hospital with a complaint of pain and swelling in the right temporomandibular joint (TMJ) area since 3 months ago. She did not have any previous history of trauma or other problems in the TMJ. Initial computed tomography scan showed destructive huge radiolucent lesion in the right TMJ without any sign and symptom of cranial nerve involvement (Fig. 1). Maximum mouth opening was 35 mm. Fine-needle aspiration showed no presence of blood or other fluid involvement. Therefore, it was decided to completely excise the lesion. Surgical treatment compromised preauricular incision and dissection with direct access to lesion after complete removal (Fig. 2). During the operation, our initial consideration concerning the lesion was to differentiate from ossifying fibroma. Frozen-section examination revealed the lesion was negative for malignancy. Subsequent to complete removal of lesion with providing hemostasis, primary closure was made. Specimen in formalin was composed of multiple fragments of creamy-white tissue with soft to rubbery consistency. The lesion was 6 × 4 × 2 cm. Microscopically, it consists of cellular tissue mainly composed of spindle cells arranged in a storiform pattern (Fig. 3), admixed C and scattered osteoclastic-like giant cell with hemosiderin-laden macrophages. In some sites, cavities with many

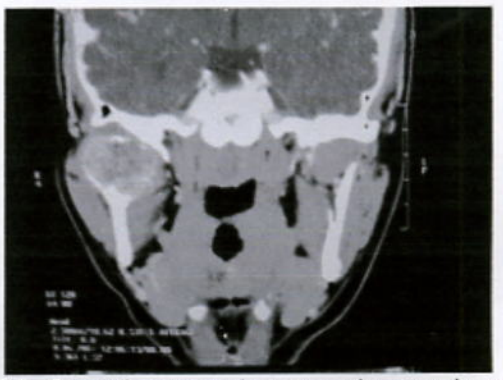

FIGURE 1. Coronal computed tomography scan shows huge radiolucent lesion in the right condyle.

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