

## MAXILLARY SOLITARY CYST: REVIEW OF LITERATURE AND CASE REPORT

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الكيس العظمي الوحيد، يعد آفة عظمية فكية غير شائعة نسبياً. وهو إحدى ما يسمى بأكياس الفك الكاذبة لأنه غير مبطن بنسيج بشري. ونظراً لاستعمال تسميات متعددة لهذه الآفة فإن ذلك يشير إلى الجدل القائم حول المنشأ المرضي لهذه الآفة. وغالباً يتواجد هذا الكيس في المنطقة الخلفية للفك السفلي ومن النادر حدوثه في الفك العلوي. وهنا نصف حالة حدوث كيس عظمي وحيد في منطقة الفك العلوي عند مريض مصاب بنقص في وظائف الدرق. وقد أظهرت الإصابة عند هذا المريض على شكل انتباج شفوي خالي من الأعراض فوق الثنايا العلوية دون أن يكون هناك سيرة مرضية تشير إلى حدوث رض موضعي سابق على هذه المنطقة. أن الهدف من هذا المقال هو الاقتراح بأن اضطرابات استقلاب الكالسيوم قد يكون عاملاً مسبباً لحدوث هذا النوع من الأكياس عند المرضى المصابين بنقص وظائف الدرق، مع توضيح الدراسات التي أجريت على هذا النوع من الأكياس التي تصيب الفك.

Solitary bone cyst (SBC), a relatively uncommon lesion of the jawbones is a type of the so-called "pseudocysts" of the jaws since the bone cavity is not lined with proper cystic membrane. The numerous synonyms coined with this lesion make it obvious that there has been much disagreement on the pathogenesis of the lesion. The SBC presents usually in the posterior region of the mandible and rarely in the maxilla. A case of anterior maxillary SBC has been presented in a patient with hypothyroidism. The lesion presented itself as an asymptomatic buccal expansion above the upper central incisors with no history of previous local trauma. This article suggests possible role of disturbed calcium metabolism as an etiologic factor for SBC in hypothyroidism patients and to present a review of the literature of the cyst.

### Introduction

The solitary bone cyst which was first described by Lucus<sup>1</sup> in 1926 is no longer considered as a rare lesion of the jaws, although it is more common in long bones, namely humerus<sup>2</sup>. In the head and neck region, the solitary bone cyst (SBC) presents commonly in the mandible,<sup>3</sup> occasionally in the maxilla,<sup>4</sup> and rarely in the zygoma.<sup>5</sup> Mandibular lesions predominate in the posterior region,<sup>2,6,7</sup>

however, lesions in the anterior mandible were reported<sup>8</sup> and found by Beasley<sup>9</sup> and Kuroi<sup>10</sup> to have an incidence of 0.13 and 0.24, respectively. In the maxilla, the lesion is relatively uncommon<sup>4,9,11-16</sup> with few reports in the literature all involving only one or two cases, with the exception of Hansen<sup>4</sup> who reported 21 maxillary cases in his series. The clinical presentation of the lesion may be asymptomatic and discovered on routine radiographic examination. The existence of presenting symptoms was reported and found to range from 3% to 35% of the cases,<sup>6,17</sup> however. These symptoms ranged from osseous expansion,<sup>6,17</sup> pain,<sup>17</sup> paraesthesia of the affected sides,<sup>18,19</sup> pathological fracture of the mandible,<sup>19,20</sup> multiplicity of the lesion,<sup>9,20-24</sup> and the presentation of SBC in the mandible and the humerus.<sup>21</sup> In addition, the

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lesion can be associated with other pathological conditions such as necrotic pulp,<sup>4,22</sup> facial cellulitis,<sup>22</sup> impacted third molar,<sup>23</sup> and odontogenic keratocyst.<sup>25</sup> The SBC was believed to have an age and sex predilection since it shows commonly between the second and third decades of life, and the lesion affects males more than the females in a ratio of 3:2.<sup>2,4</sup> Kougars and Cale,<sup>26</sup> on reviewing 161 cases of SBCs, found that there is no gender predilection for the lesion, however. The diagnosis of SBC is mainly made on clinical basis, i.e., by surgical intervention. The exploration procedure reveals a relatively empty bone cavity with little tissues, connective tissue,<sup>2</sup> with either little fluid content or gas in the cavity. The fluid content of the cyst was said to have a higher bilirubin level<sup>27</sup> and/or acid phosphatase level<sup>28</sup> as compared to other cyst fluids. Histologically, the SBC is characterized by the absence of an epithelial lining with only loose connective tissue covering the bony wall<sup>2,6,29</sup> containing congested capillary vessel, extra-vascular blood cells, hemosiderin and multinucleated giant cells.<sup>30,31</sup> Radiographically, the SBC commonly presents as a single, unilocular, well demarcated, radiolucency of variable size. In the premolar region, the lesions may demonstrate a scalloped appearance due to the projection of bony cavities into the intraradicular septa.<sup>31</sup> The involved teeth may be displaced, but seldomly resorbed and often reposition themselves after resolution. Atypical radiographic representation of the SBC has been reported by Mitchell and Ward-Booth<sup>32</sup> as a multiloculated radiolucency, however.

Various treatment modalities were adopted for SBC: (1) keeping the case under observation and waiting for spontaneous regression;<sup>2,4,6,7,33</sup> (2) aspiration of the contents of the SBC;<sup>34</sup> (3) surgical exploration and curettage to stimulate bleeding, healing, and initially to confirm the diagnosis;<sup>16</sup> (4) packing with gel foam saturated with thrombin and penicillin;<sup>35</sup> (5) endodontic intervention;<sup>36,37</sup> (6) injection of methyl prednisolone acetate (MPA) solution for treatment of long bone cases;<sup>38</sup> (7) injection of autogenous blood to stimulate the osteogenic activity;<sup>39</sup> and (8) bone grafting.<sup>40</sup> Recurrence of the lesion is not commonly encountered, however, it is being postulated that existence of another cyst within the bone cavity, which may not have been enucleated, is a cause for recurrence.<sup>32,40,41</sup>

### Case Report

On April 6, 1986, a 33-year-old Syrian woman was referred to the Oral Surgery Unit, College of Dentistry, King Saud University in Riyadh, Saudi Arabia for evaluation of an asymptomatic unilocular radiolucency related to the apices of the maxillary central incisors, which was discovered on routine radiographic screening.

The patient gave a history of hypothyroidism in the last 10 years and under treatment with Eltroxin 200 mg tab. once a day. In addition, there was a history of calcium malabsorption especially during pregnancy. No history of local trauma was reported.

On examination, the teeth were vital and they responded normally to both thermal and electrical pulp testing. There was no periodontitis in the area and the teeth were not mobile or tender to percussion. On palpation, there was a non-tender osseous expansion of the labial cortical plate of bone about 2 cm in diameter above the maxillary central incisors. The overlying mucosa was intact and of normal appearance. Clinically, no draining sinuses or lymph node involvement were observed [Fig. 1]. Radiographic examination showed a 1.5 x 0.5 cm. radiolucent area with thin sclerotic border superior to the apices of the maxillary central incisors. The apical third of the roots of the central incisors were involved in the radiolucency. The lamina dura of the teeth was intact [Fig. 2].

Complete skeletal survey of the patient revealed an irregular contour of the iliac crest. Hematological study showed slight poikilocytosis and polychromasia of the RBCs and an elevated alkaline phosphatase level at 314 U.I. Serum calcium and inorganic phosphorus were within normal limits. The differential diagnoses included SBCs, nasopalatine duct cyst, primordial cyst, central giant cell lesion, early stage of fibro-osseous lesion, odontogenic tumor and central hemangioma.

On April 13, 1986, the decision was made to explore the area under local anaesthesia using two carpules of 1.8 ml of 2% plain xylocaine solution after initial failure of aspiration biopsy.

A semilunar mucoperiosteal flap was raised from the right to the left lateral incisors exposing the bony expansion labially. A window was opened in the expanded labial cortical plate using the postage



Figure 1. Preoperative view showing intact mucosa and normal colored teeth.

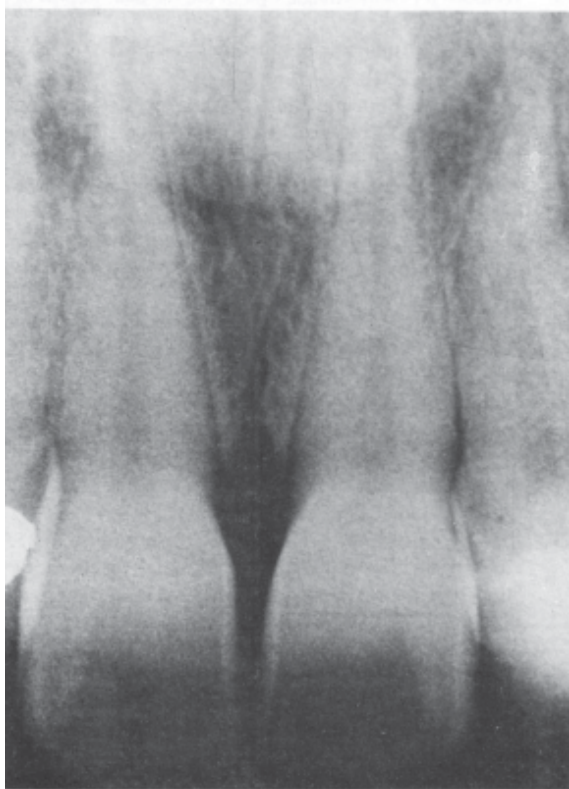


Figure 2. Preoperative radiograph.

stamp bur technique. This revealed a bony cavity with very little blood-tinged fluid and some fine friable soft tissue [Fig. 3]. Curettage of the cavity was performed and the flap was sutured back with black silk suture. The removed pieces of bone, with the minimal amount of soft tissue and fluid, were sent for histopathological study. The wound healed uneventfully and the stitches were removed after seven days of the operation. Two weeks after the



Figure 3. Operative view showing empty bone cavity.

surgery, endodontic treatment of the maxillary right central incisor was started after the tooth showed diminished vitality response.

Histopathological report confirmed the clinical diagnosis of SBC made from clinical examination and surgical intervention.

#### Discussion

The numerous synonyms coined with the SBC which varies from progressive bone cyst, traumatic bone cyst, traumatic hemorrhagic cyst, hemorrhagic cyst, extravasation cyst, hemorrhagic extravasation cyst, unicameral cyst, simple bone cyst reveals a considerable disagreement regarding the pathogenesis of the lesion. The pathogenesis and the etiology of SBC still remains uncertain, with the most widely accepted theory of an intra-medullary hemorrhage as a result of trauma, which fails to organize, and the subsequent degeneration of the clot producing an empty cavity within the bone.<sup>2-4,13,15,16</sup> On the other hand Kougars and Cale<sup>26</sup> found that in 161 reviewed cases, the history of prior trauma among these patients was equivalent to that described for the general population. Other theories for the pathogenesis of SBC<sup>2,9,17</sup> included : (1) infection of bone marrow; (2) loss of blood supply to a hemangioma or lymphoma; (3) cystic degeneration of existing bone tumor; (4) changes and reduction in the osteogenic activity; (5) faulty calcium metabolism as a result of systemic disease, such as parathyroid diseases; (6) ischemic necrosis of the fatty bone marrow; (7) low grade chronic infection; (8) imbalance between the osteoclastic and osteoblastic activity due to trauma; (9) developmental defect; (10) failure of mesenchymal

tissue to form bone and cartilage, and instead becomes immature as multiple bursa-like synovial cavities.<sup>41</sup>

The present case suggests the possible implication of hypothyroidism which induced disturbed calcium metabolism in the pathogenesis of SBC without denying the role of trauma in some cases as an initiating factor for their lesion. There is also the remote possibility of drug-induced (Eltroxin) etiology in this case.

Hosseini<sup>41</sup> suggested that according to existing theories of pathogenesis, the SBC should appear with the same frequency in the maxilla as in the mandible. Hansen<sup>4</sup> and Kaffe et al<sup>43</sup> assumed that the low incidence of maxillary SBC is due to the superimposition of various anatomical radiolucencies in the maxilla which obscures the SBC. This report further supports the latter view specially for anterior maxillary SBC, where suspect of superimposition of the naris can cause some cases of SBC to be missed specially if the lesion is asymptomatic.

The diagnosis of SBC prior to surgical intervention, constitutes in most instances a great difficulty to general dental practitioners as the lesion can have different presentation in nearly every region of the jaws. For these reasons, Rushton<sup>29</sup> adopted the following criteria for establishing diagnosis : (1) a single lesion, (2) no epithelial lining, (3) no infection, (4) no perforation of the bony walls, and (5) fluids in the lesion. Later on, Hansen<sup>4</sup> modified these criteria as follows : (1) upon surgery, the lesion is essentially empty and, occasionally, the cavity contain some fluid and/or small amount of tissues; (2) other findings (clinical, radiographic, historic, histopathologic, etc.) do not exclude the diagnosis of SBC. Compared to both Rushton<sup>29</sup> and Hansen<sup>4</sup>, the reported case was found to meet all the described criteria.

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