Case Reports

ODONTOGENIC KERATOCYST: A CASE REPORT IN A FIVE-YEAR-OLD SAUDI BOY

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فيليبسين عام ١٩٥٦م وصف الكيس السني المتقرن وظلت هذه الآفة تثير اهتمام الأطباء بسبب النمو الطبيعي لهذه الآفة وميلها إلى الشفاء بعد الاستئصال الجراحي. وبسبب سلوكية هذه الآفة، فإن التشخيص الصحيح لهذه الآفة قبل العلاج ضروري للتأكد من أن المعالجة الجراحية ضرورية. يحدث الكيس السني المتقرن في مختلف الأعمار إلا أن حدوثه يكثر في العقد الثاني والثالث من العمر. ويصيب الذكور أكثر من الإناث وتمتد الآفة الكيسية بشكل البالون وتتوسع بالاتجاه الأمامي الخلفي. وشعاعيا يظهر بشكل جوف أو تجاويف صغيرة متعددة ناقلة للأشعة لها حواف متصلبة وواضحة.

الهدف من هذه الدراسة إظهار النواحي السريرية والشعاعية للكيس السني المقترن عند مريض يبلغ من العمر خمسة سنوات أتى إلى كلية طب الأسنان بجامعة الملك سعود يشكو من انتباج شديد في الناحية اليسرى من الذقن أدى إلى عدم تناظر في جانبي الوجه. وأخبرنا والده أن الانتباج لوحظ منذ ستة أسابيع فقط وازداد تدريجيا بالحجم دون أية آلام مرافقة. والأسنان المجاورة له أصيبت بالتقلقل حيث الحركة فيها من الصنف الثاني والثالث. تمت معالجة الحالة بطريقة محافظة بفتح الكيس وسحب السائل الكيسي وتسكيره بشكل جزئي كمرحلة ثانية لإتمام المعالجة. ولوحظ تشكل وتوضع العظم على جدران الكيس بعد ستة أشهر من المعالجة الجراحية.

An odontogenic keratocyst possesses tumor-like characteristics because of its clinical behavior. The aggressive growth, high recurrence rate and seeding of the cyst into soft tissue exemplify this behavior, ft has been reported that the age distribution for keratocysts has a biomodal characteristic, with the first and greater peak in the second and third decades and a second lower peak in the fifth decade. Incidence of occurrence before age ten is low. In this report, odontogenic keratocyst in a five-year-old Saudi boy is presented and the management of the case is briefly discussed.

Introduction

Ever since Philipsen[1] described the odontogenic keratocyst in 1956, the lesion has continued to raise a considerable clinical interest because of its unusual growth pattern and tendency to recur after surgical removal.[2-5] In view of this behavior, a precise pre-operative diagnosis of the lesion is necessary to assure a proper surgical approach and successful eradication.[6-8]

Odontogenic keratocysts occur over a wide age range with a pronounced peak incidence in the second and third decades.-[3,7,9-13] It is seen more frequently in males rather than in females.[7,11] The cystic lesion expands in a unicentric ballooning pattern and enlarges predominantly in an anterior-posterior direction. Radiographically, it appears as unilocular or multilocular radiolucencies with well-defined sclerotic margin. The growth of odontogenic keratocyst may occur rapidly in young children usually preventing teeth eruption and mimicking often a dentigerous cyst.[8]

The purpose of this article is to present the clinical and radiographical features of odontogenic keratocyst in a five-year-old patient. A brief discussion concerning the management of this case is also included.

Received 30/08/92; revised 29/12/92; accepted 03/03/93

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Case Report

A five-year-old boy reported in January 1992 to the Orai Surgery Clinic at the College of Dentistry, King Saud University in Riyadh with a chief complaint of swelling on the left side of the chin that caused a considerable asymmetry of the face [Fig. 1]. As narrated by his father, the swelling was noted six weeks previously while its size gradually, but progressively, increased. It was painless and caused no alteration in sensation over the mental nerve distribution. The labio-buccal sulcus was obliterated by a bony swelling and fluctuation was elicited in its center [Fig. 2], Bony expansion extended lingually and the overlying mucosa showed normal texture. The adjacent teeth were all mobile and were assigned grade II to grade III mobility. Radiographically, a well-circumscribed radiolucency extended from the left second deciduous molar to the right deciduous canine involving almost the whole height of the mandible [Fig. 3] and extended to a greater extent labiobucally [Fig. 4]. The erupting permanent teeth were displaced from their path of eruption and

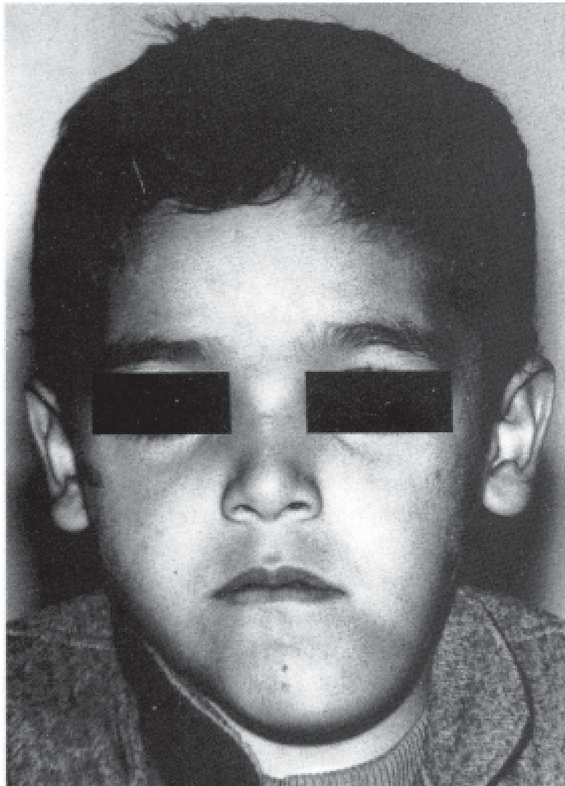

Figure 1. Anterior view showing facial asymmetry.

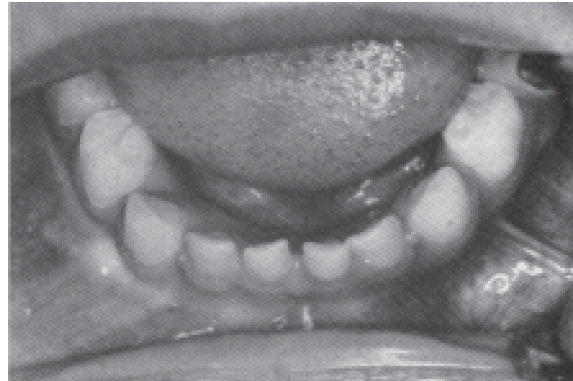

Figure 2. Intraoral view showing the obliterating left labio-buccal sulcus.

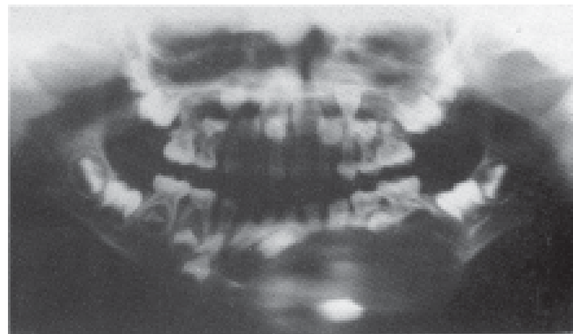

Figure 3. An orthopantomogram demonstrating a cyst occupying almost the whole height of the mandible.

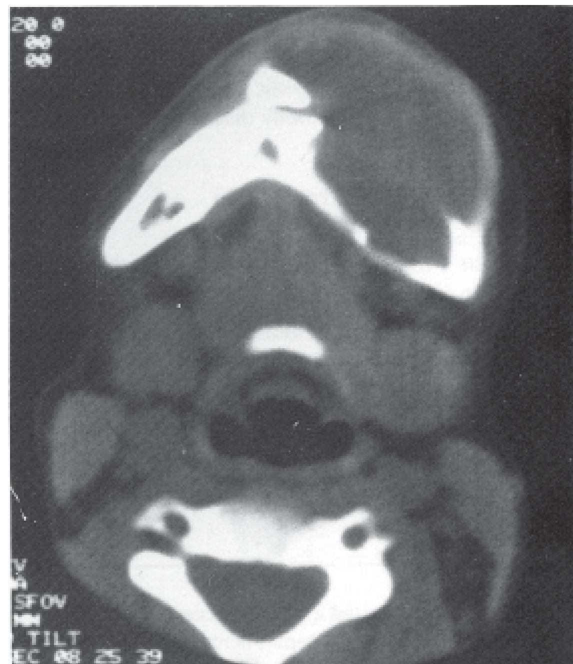

Figure 4, CT scan of the mandible showing bucco-labial extension of the cystic lesion.

pathological resorption of the related deciduous teeth was evident radiographically. Body scanning and physical examination ruled out the existence of a basal-cell nevus syndrome. The cyst content was aspirated and was of thick pus-like consistency. Electrophoresis of the cystic fluid demonstrated low soluble protein content (< 3.75 g./lOO ml). The tentative diagnosis of odontogenic keratocyst was made and confirmed afterwards by histological examination of the surgical specimen. The cross section showed parakeratinized stratified squamous epithelium with uniform thickness in most areas, and with a prominent palsading basal layer, which was consistent with the pre-operative diagnosis of odontogenic keratocyst [Fig. 5].

The case was managed conservatively by decompression of the cystic cavity, followed six months later by enucleation and primary closure, as a second stage procedure, in order to complete the treatment. The decision of the second stage operation was made upon the deposition of bone around the cystic wall and the steady diminishing in cystic cavity size. Eleven months later, the osseous defect was completely filled in with normal bone [Fig. 6].

Figure 5. Histological section illustrating the lining of parakeratinized stratified squamous epithelium with a prominent balsading basal layer (magnification x 40; H & E stain).

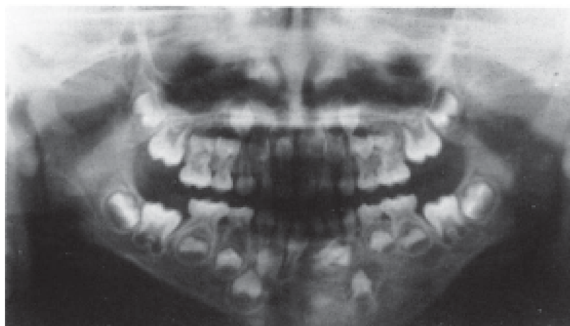

Figure 6. Panoramic radiograph showing complete obliteration of the cystic cavity by normal bone, eleven months post-operatively.

Discussion

Previous reports[7-13] indicated that the incidence of odontogenic keratocyst covers wide age ranges of seven to eighty-three years (Table 1). The age of the patient was less than what had been reported previously. Rapid growth in this case might be explained by the aggressive nature of this lesion as reported earlier, particularly in young children.[8]

Various modes of therapy have been advocated for the treatment of keratocyst and these include marsupialization, enucleation and primary closure, enucleation with excision of the overlying mucosa and packing, decompression with secondary enucleation, treatment of the whole cyst cavity with Carnoy's solution prior to enucleation and radical surgery. In 1970, Browne[7] found no significant differences in recurrence among these modalities but a recurrence rate of 0.0% with a mean follow-up of 7.4 years in a group of patients treated with Carnoy's solution as cauterizing fixative agent,[14] was reported.

Forssell and his coworkers[15] found that the surgical difficulties in removing the thin friable capsule and the existence of bony perforation that is often associated with soft tissue adherence are the likely possible factors responsible for recurrence. Nevertheless, their observation regarding the high occurrence of recurring keratocyst in younger patients was not confirmed by Vedtofte and Praetorius.[10]

Marsupialization is not the accepted treatment for odontogenic keratocyst because pathological epithelium is left *in situ*. This modality, however, remains the treatment of choice for large unilocular cyst with extremely thin lining, provided it is followed by a second stage operation consisting of enucleation with primary closure or packing.[4] The simple conservative treatment employed in this case was deliberate and regarded as the first stage of management. It was instituted primarily to prevent the likely pathological fracture of the jaw

Table 1. Incidence of occurrences for odontogenic keratocyst.

Author	Year	No. of Patients	Sex M	Sex F	Age Bracket	Peak incidence/ Decades
Browne[7]	1970	65	38	26	8-83	II and III
Payne[9]	1972	81	38	40	7-74	II
Raddenand Reade[11]	1973	66	44	22	8-81	II and III
Vedtofteand Praetorius[10]	1979	72	38	34	10-71	II and V
Partridge and Towers[8]	1987	60			11-81	II
Angelopoulos and Nicolatou[12]	1990	79	58	21	10-80	V and VI
Narty and Saini[13]	1990	10	7	3	25-72	II and III

consequent to enucleation and to permit eruption of teeth involved in the cystic process. The second stage procedure was undertaken when the cystic cavity was eventually obliterated by the filling of bone, permitting teeth to move up to where it could be aligned easily by orthodontic appliance.

A long post-operative follow-up over the years may demonstrate the recurrence to be less dramatic. Therefore, an observation at regular interval for a period of at least ten years is found to be very necessary to make a diagnosis of recurrence without delay.[7,15]

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