So-called simple bone cyst of the jaw: A family of pseudocysts of diverse nature and etiology

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Abstract: The nature and etiology of so-called simple bone cyst (SBC) are still a subject of debate. Our comprehensive review of the literature suggests that SBC, which appears histologically to be a single entity, has different natures and etiologies, resulting in divergent clinical features. In addition, an interesting case of mandibular SBC in an 11-year-old girl is presented with details of radiographic changes over a 7-year period. Fully documented patient records revealed that this lesion originated in the apical area of the first molar and took about 4 years to develop into a clinically evident bony expansion. (J. Oral Sci. 41, 93-98, 1999)

Key words: hemorrhage; pathogenesis; pseudocyst; simple bone cyst; time-related radiographic change; trauma.

Introduction

Despite the extensive volume of literature on so-called simple bone cyst (SBC) that has accumulated during the 70 years since Lucas (1) first reported the condition, there is still little information about its precise nature and etiology. Since the vast majority of SBCs produce no symptoms and can remain undiagnosed for many years, they are often fully developed at the time of diagnosis. It is therefore difficult to obtain data at an earlier stage of their development.

We encountered a case of mandibular SBC for which monitoring was possible over a 7-year period. This case was considered very useful for helping to clarify not only the early stage and the rate of growth, but also the pathogenesis of SBC. The purpose of this article is to characterize the entire nature of SBC on the basis of this case, and a comprehensive review of the English literature (1-111).

Case report

A 4-year-old girl was first seen in October 1990 for treatment of dental caries. Results of radiographic examinations were essentially within normal limits (Fig. 1 A), and over the next two years, no significant radiographic changes were evident. In May 1993, she was admitted again for routine dental treatment. A panoramic radiograph showed an ill-defined radiolucency with loss of trabeculae in the periapical area of a vital standing first molar in the right mandible. The sclerotic margin of the dental follicle around the developing permanent second premolar was partially interrupted (Fig. 1 B). There was no history of trauma or infection. Although it was requested that she return for reevaluation periodically, the patient was not seen again until June 1995, when a panoramic radiograph revealed that the radiolucency had increased in size (Fig. 1 C). Again, follow-up was unfortunately interrupted until June 1997, when the patient was readmitted with a painless swelling on the buccal aspect of the premolar region, although the involved teeth had erupted in a functional position. Radiographs showed a well-demarcated, unilocular cystic lesion with scalloped margins (Figs. 1 D and 2). Under a working diagnosis of developmental odontogenic cyst, a biopsy was obtained, and this confirmed the presence of SBC. At the time of decompression, the cystic cavity was empty. Since then, radiographic examinations have revealed marked reduction of radiolucency. The patient will be closely followed upon a long-term basis.

Discussion

SBC is an intraosseous pseudocyst having a tenuous lining of fibromyxomatous tissue without an epithelial component (112-114). It is not a common lesion (112,114,115). The nature and etiology are still far from being established conclusively, and this is reflected in the variety of terms used to describe the condition, including traumatic, hemorrhagic or solitary bone cyst, progressive or idiopathic bone cavity, extravasation cyst and combinations of these synonyms (1-111). At present, the name SBC, proposed by Bernier and Johnson (18), seems to be the most appropriate for representing the overall nature of the entity.

It is generally conceded that great variation is evident in the clinical features of SBC (55, 62, 68, 79, 100, 101, 108, 111). Aside from classic cases diagnosed during the second decade of life, the age of patient ranged from 2 (8) to 75 (62) years. The majority of lesions were asymptomatic, being detected accidentally by radiographic examinations carried out for other reasons, although pain, swelling, paresthesia, displacement and root resorption of the involved teeth, fistula and pathologic

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fracture have also been reported (35, 53, 56, 62, 67, 68, 78, 100, 101). The lesion was usually solitary and located in the dentulous area of the mandible. Unusual locations reported included the condylar process (88, 98, 105, 106) and zygomatic arch (86). Bilateral or multiple lesions were described in 4-10% of cases (44, 71, 76, 78, 81, 82, 85, 98, 100, 103). Although spontaneous resolution has been well-documented (40, 84, 107), recurrent lesions have also been described (39, 63, 64, 77, 94, 95, 101, 108, 111). Therefore, so-called SBC has two distinct patterns of clinical behavior: solitary, asymptomatic, self-limiting lesions with a tendency for spontaneous healing, and solitary or multiple, progressive lesions with a potential for recurrence. Moreover, SBC is sometimes found in association with various non-neoplastic bone lesions, including giant cell reparative granuloma (56), aneurysmal bone cyst (74) and fibro-cemento-osseous dysplasia (116-122). Thus, it is conceivable that there are three different forms of SBC.

A few radiographic follow-up studies of SBC have been published (17, 33, 77, 97), but these have not satisfactorily clarified the details of early onset. In the present case, the radiographic evidence indicate that the lesion was apparently of endosteal origin, primarily involving the medullary bone with secondary involvement of the cortical bone. For technical reasons, although it was not possible to accurately compare the panoramic films with respect to the size of the radiolucency, the site of origin was the periapical area of the mesial root of the first molar. It took about four years to produce bony expansion. One previously unreported feature of this case was that disappearance of the dental follicle around the permanent second premolar was evident during development of the lesion. The pure cystic nature of this lesion, and the minimal pressure

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Fig. 1 Comparative panoramic radiographs over 7-year period: A) 1990 (4 years old); no radiographic bone changes in the mandible. B) 1993 (7 years old); small ill-defined radiolucency in a periapical area of right mandibular first molar (arrow head); note discontinuity of a sclerotic margin of dental follicle of erupting second premolar. C) 1995 (9 years old); well-defined radiolucent lesion with erosion of lower border of mandible (arrow head); note disappearance of follicular space of second premolar. D) 1997 (11 years old); more pronounced radiolucency extending from first premolar to mesial aspect of first molar (arrow head); note extremely erosion of lower cortical border. Second premolar erupted into normal position.

Fig. 2 A periapical radiograph taken in 1997. Scalloped radiolucent lesion in the right mandible; lamina dura of first molar appears thin but distinct. No apical root resorption is evident.
it produces may allow the normal dentition to develop. This is, however, not a constant finding (33).

To date, numerous theories about the etiology of SBC have been proposed (1-111). These include sequelae of traumatic intramedullary hemorrhage, cystic degeneration of benign neoplastic lesions, low-grade infection, faulty calcium metabolism, a local disturbance of bone growth, ischemic marrow necrosis, venous obstruction and localized alteration of bone metabolism resulting in osteolysis. However, no theories have been completely proved. The most widely accepted traumatic-hemorrhagic theory fails to explain adequately the origin of the present lesion. The differing clinical features suggest that so-called SBC may not have a single common cause.

In summary, although the present case cannot elucidate the pathogenesis of SBC, some insight may be gained by further radiographic follow-up.

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