Intraosseous cystic lesions of the jaws in children: A retrospective analysis of 47 consecutive cases

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Objectives. The aim was to review the characteristics and treatment approach in 47 cases of intraosseous cystic lesions of the jaws in children.
Study design: Forty-three patients, 2-14 years old, with 47 cystic lesions of the jaws, were treated during the period 2000-2007. All cysts were surgically removed. No bone grafts were used and the teeth involved were preserved whenever possible. Removed tissues were examined histologically. Follow-up period ranged from 6 months to 7 years.
Results. In 74.5% of the cases, cysts were enucleated, and in 17.0% they were marsupialized. Most commonly, the cysts were dentigerous (20, 42.6%), followed by eruption cysts, odontogenic keratocysts, and radicular cysts (10.6% each) and buccal bifurcation cysts (8.6%).
Conclusion. Cystic lesions in children were found to be mainly of developmental origin. Treatment was surgical removal without interfering, when possible, with the development of the dentition. Surgical approach was usually enucleation and, to a lesser extent, marsupialization. **(Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2009;107: 485-492)**

Cystic lesions of the jawbones have been recognized, described, and studied in numerous publications, mostly in adults and to a lesser extent in children, where they are usually found in the literature as case reports.

Differences may be found among authors regarding their classification[1-4] and their terminology.[2,5-7] Radicular cysts of inflammatory origin are by far the most common cysts in the general population[3,5]; this is not the case in children and adolescents, where dentigerous cysts and other cystic types of developmental origin most often develop.[7-9]

Dentigerous cysts, associated with an unerupted tooth, appear most often during the second or third decade of life, with a peak incidence during the teenage years.[3,7,10] They frequently develop in the posterior area of the mandible, followed by the maxillary canine region.[10,11] Other types of cystic lesions, such as eruption and buccal bifurcation cysts, are also seen, usually only in children.[2,6,10,12,13] Additionally, odontogenic keratocysts, recently reclassified by the World Health Organization as keratocystic odontogenic tumours,[14] have been reported to have a peak incidence in the second and third decades of life,[7] although other studies have found them mostly in the fourth and fifth decades.[2,15] Finally, aneurysmal bone cysts or cavities generally affect adolescents.[7,16-18]

Diagnosis of cystic lesions is based on clinical and radiologic findings. Clinically, they may be asymptomatic or may have acute or chronic findings. In the latter case, swelling and pain may be the presenting symptom. Their size and location are evaluated by routine radiographs, computerized tomography (CT), and 3-dimensional(3D) CT.[19] Fine-needle aspiration and biopsies may assist the diagnosis in selected cases.[20,21]

Surgical removal is the treatment of choice, which, depending on the case, may be enucleation, marsupialization, curettage, removal of the content with curettage, or, occasionally, surgical resection.[3,5,10]

Histologic examination usually clarifies the type and origin of the cystic lesion and helps the clinician to decide the follow-up period of the patient.

The aim of the present retrospective study was to review and analyze the characteristics and the surgical approach in 47 consecutive cases of intraosseous cystic lesions of the jaws in children treated in our clinic during an 8-year period.

PATIENTS AND METHODS

Forty-seven jaw cysts in 43 young patients aged 2-14 years were treated from January 2000 to December

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2007. Patients were referred for consultation, usually for swelling and pain, from general dental practitioners, pediatric dentists, and pediatricians.

After clinical examination, panoramic, periapical, and occlusal radiographs were taken and, in some cases of large (>4 cm in diameter) cystic lesions, additional radiographs including CTs or 3D CTs were prescribed. Diagnostic biopsy was performed in cases with doubtful diagnosis in lesions >3 cm.

All cystic lesions were treated surgically under general anesthesia with an intraoral approach in the majority of the cases (46, 97.9%). An extraoral approach was performed once for removal of the contents of a large aneurysmal cavity of the mandibular angle (12 cm in diameter; Fig. 1).

Regarding the surgical approach, enucleation in 1 stage (Fig. 2) or marsupialization were chosen accordingly, the latter used only in eruption cysts and in cases with large cystic dimensions. When marsupialization was performed, the cavity was lightly packed with Fucidine gauze changed twice a week for a period of 2-3 weeks (Fig. 3). No bone grafts were used to fill the cavities. Involved permanent teeth were preserved whenever possible. Radicular cysts were enucleated with simultaneous apicectomy and retrograde filling of the causative teeth (Fig. 4). Mucoceles of the maxillary antrum were removed through a window of the anterior wall, which was then repositioned and stabilized with resorbable osteosynthesis material. Aneurysmal bone cavities, after biopsy, were treated with complete removal of their content and curettage, and the simple bone cavity was treated with curettage only, until bleeding occurred.

Removed tissue material was always examined histologically. The follow-up period ranged from 6 months to 6 years. Patients were discharged when both clinical and radiologic examination showed complete regeneration of the bone in the affected areas, with no signs of recurrence.

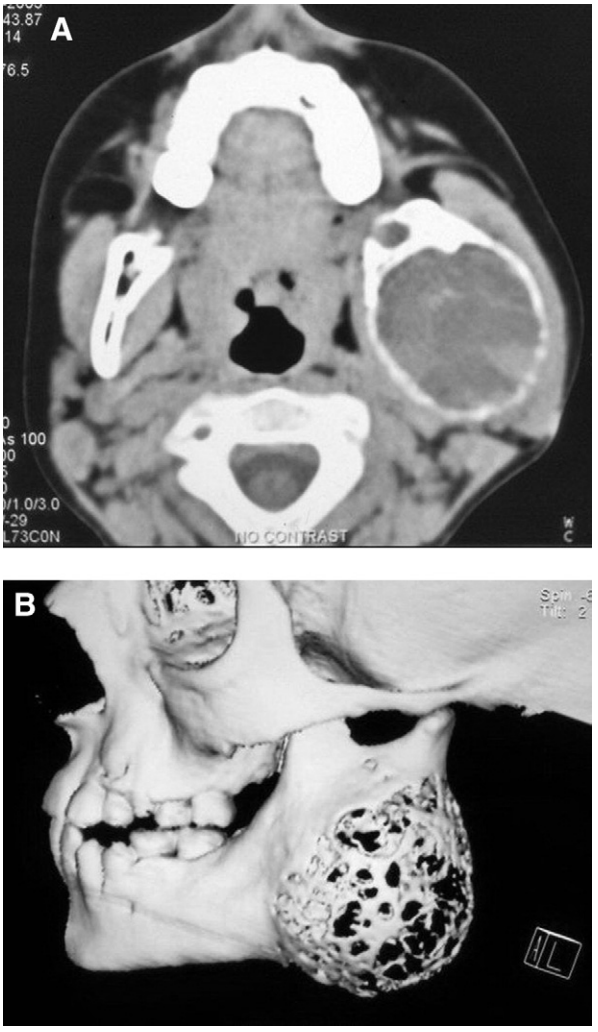

Fig. 1. Large aneurysmal bone cavity in a 10-year-old boy. **A,** Computerized tomography (CT) showing the destructive lesion at the left mandibular ramus. **B,** Three-dimensional CT presenting the expansion and partial destruction of the cortex of the angle and the ramus.

RESULTS

All data of the patients are summarized in Table I. Boys were more frequently affected (28, 58.5%) than girls (19, 41.5%). The mean age of our patients was 9.95 ± 2.8 years. When each type of cystic lesion was examined separately, cases of eruption cysts had a mean age of 7.4 ± 3.9 years, buccal bifurcation cysts 8.0 ± 0.8 years, aneurysmal bone cavities 10 ± 3 years, dentigerous cysts 10.2 ± 2.8 years, radicular cysts 10.5 ± 2.5 years, and odontogenic keratocysts 11.6 ± 1.5 years. In all cases, cystic lesions were more frequently found in the mandible (29 cases, 61.7%).

Regarding the presenting symptoms and signs, 44 cases revealed intraoral swelling, 12 cases extraoral swelling, 10 cases local pain, 2 cases presence of a fistula, and 1 case of granulomatous tissue.

Diagnostic biopsy was performed before operation in 4 cases: in 2 cases of odontogenic keratocysts and in 2 cases of aneurysmal cysts.

The majority of cystic lesions were dentigerous (20, 42.6%), followed by eruption cysts (5, 10.6%), odontogenic keratocysts (5, 10.6%), radicular cysts (5, 10.6%), buccal bifurcation cysts (4, 8.6%), aneurysmal bone cavities (3, 6.4%), and mucosal cysts of the maxillary antrum (2, 4.3%). Nasopalatine, simple bone cavity, and midline cysts, were found in 1 case each (Table I). Regarding the most frequently found dentigerous cysts, 13 of them (65.5%) were located in the posterior

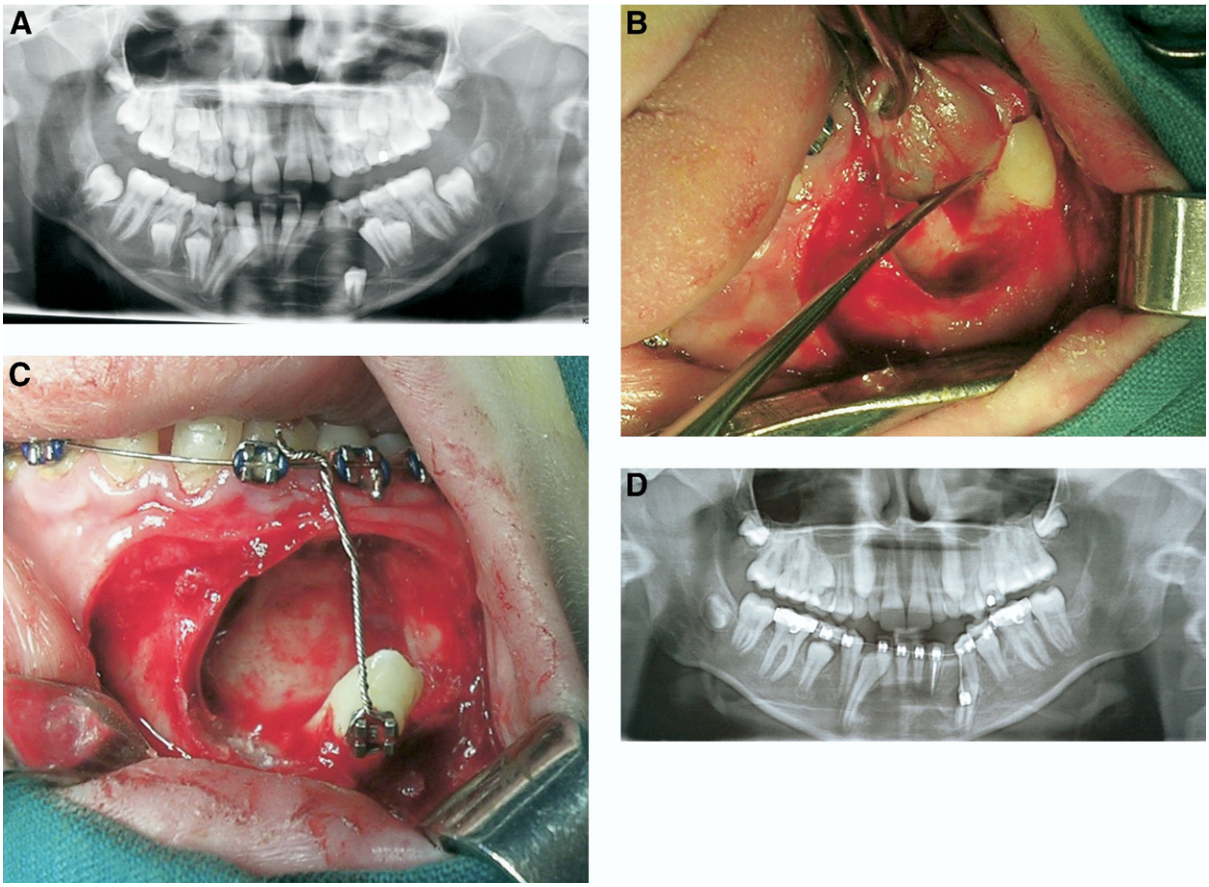

Fig. 2. Dentigerous cyst of lower left canine in a 12-year-old boy. **A,** Orthopantomogram (OPG), showing the radiolucent lesion around the impacted lower left canine being transpositioned at the lower mandibular border. **B,** Enucleation of the cyst. **C,** Immediate placement of orthodontic appliance. **D,** OPG 1 year after surgery, showing bone regeneration and the canine being almost erupted in an upright position.

region of the jaws, 6 (30.0%) in the canine region, and 1 (5%) associated with an impacted mesiodens; in 2 cases (nos. 29 and 30), the patients had >1 cyst each (Fig. 5).

Regarding the surgical approach, enucleation was performed in 35 cases (74.5%), marsupialization in 8 cases (17.0%), removal of the contents of the cystic cavity followed by curettage in 3 cases (6.4%), and curettage only in 1 case (2.1%) (Table I). More specifically 18 dentigerous cysts were enucleated and 2 larger ones were marsupialized (Figs. 2 and 3). In these cases, when located in the anterior region involved incisors were preserved and followed up radiologically, and in the posterior region involved molars, premolars, and the mesiodens were removed together with the cyst.

One odontogenic keratocyst was treated with removal of its content and curettage, and another large one was marsupialized first and removed later. The latter lesion belonged to a patient (no. 11) who was diagnosed to suffer from basal cell nevus syndrome, presenting with additional basal cell carcinomas of the face.

Three of the buccal bifurcation cysts were enucleated together with the involved first molar, and in the fourth case the tooth was preserved.

Intraoperatively, the only notable complication was a rather intense hemorrhage in the case of a large aneurysmal bone cavity (patient no. 14), which was controlled with local hemostatics.

Postoperative healing of all patients was uneventful. There was no evidence of disease recurrence in 40 out of 43 patients during the follow-up period. There was 1 recurrence in a case of odontogenic keratocyst 3 years after its removal (patient no. 11). Two more patients developed cystic lesions in different locations of the jaws. In detail, in a case of buccal bifurcation cyst, 2 years after the initial removal, patient no. 19 presented with a large radicular cyst at the apex region of a

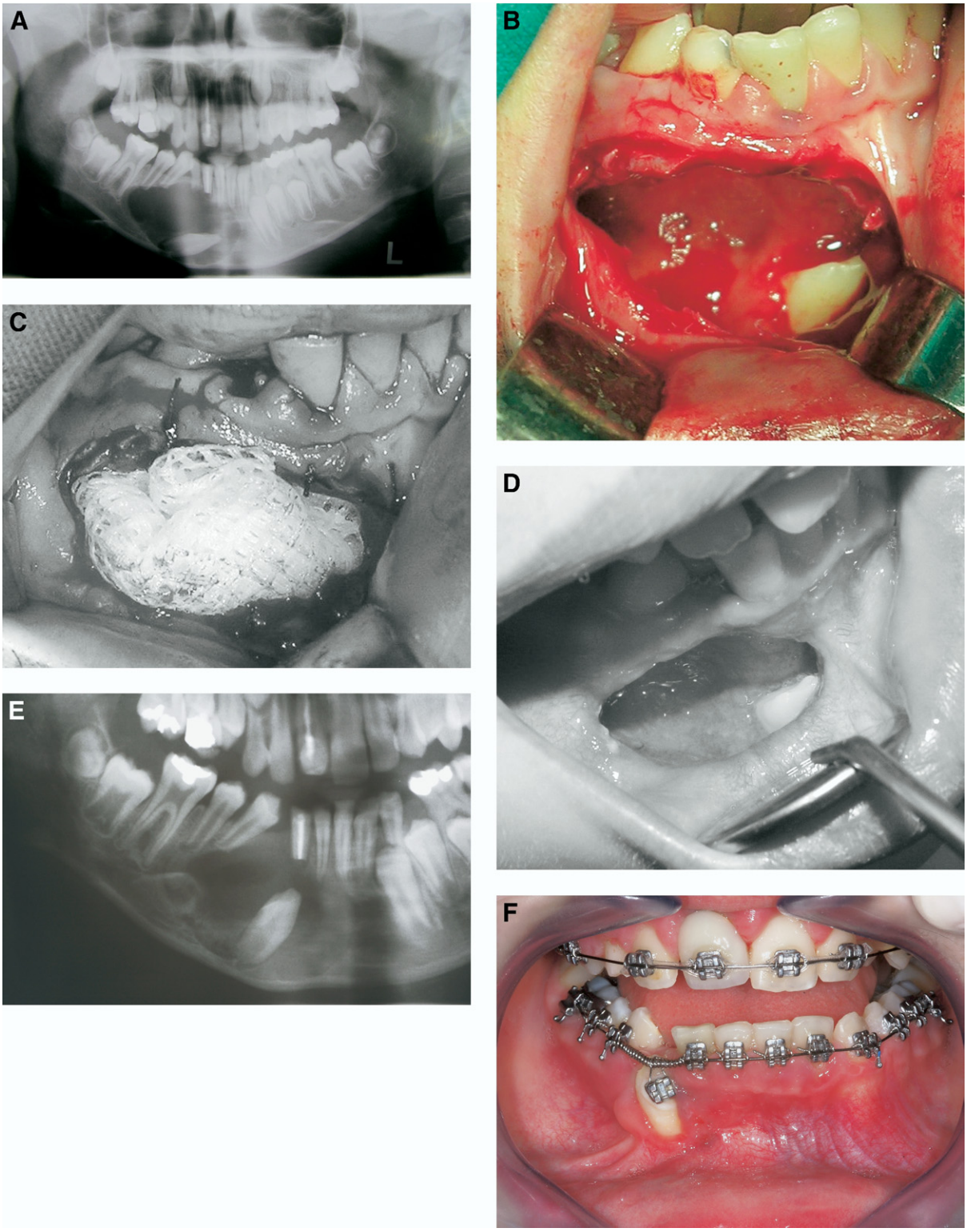

Fig. 3. Dentigerous cyst of the lower right canine in an 11-year-old boy. **A,** Orthopantomogram (OPG) showing the radiolucent lesion around the impacted lower left canine being transpositioned at the lower mandibular border. **B,** Marsupialization of the cyst. **C,** Placement of fucidine gauze. **D,** Postoperative view after 6 months. **E,** OPG showing bone regeneration and the canine being almost erupted in an upright position. **F,** Orthodontic treatment.

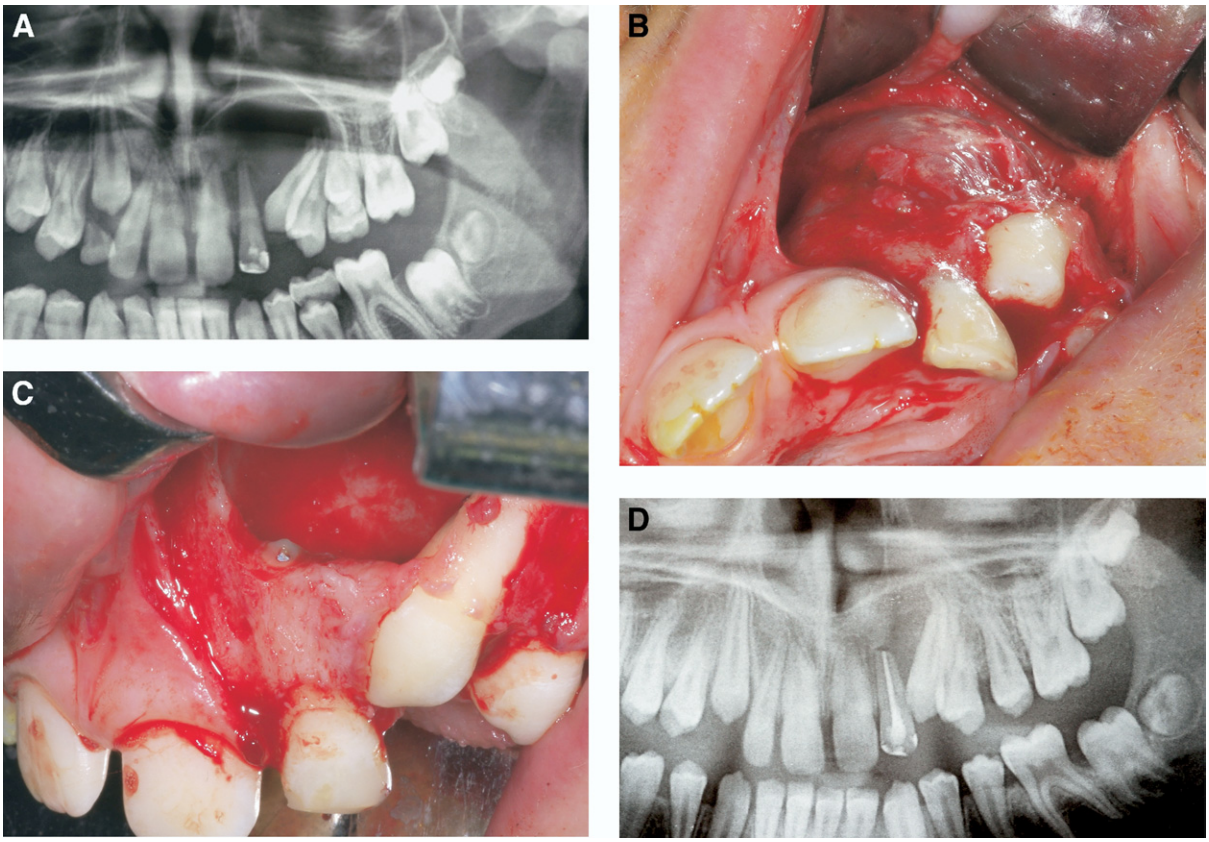

Fig. 4. Periapical cyst of the left lateral upper incisor in a 12-year-old boy. **A,** Orthopantomogram (OPG) showing the radiolucent lesion. **B,** Intraoperative view of the cyst. **C,** Apicectomy and retrograde filling of the causative tooth. **D,** Postoperative OPG after 1 year.

maxillary canine, and in a case of a dentigerous cyst of the anterior mandible, 2 years after the initial removal, patient no. 30 developed a new one at the mandibular angle. Both patients were reoperated.

DISCUSSION

The present study evaluated the results of a retrospective analysis of jaw cysts in a definitive child population up to 14 years of age. A male predominance was observed (male-female ratio 1.47:1), in agreement with findings of earlier studies in both adult and child populations.[5,9] In contrast, Bodner[8] found an almost 1:1 ratio in children.

The initial appearance in the majority of cystic lesions of developmental origin occur during childhood and are associated with continuous growing and subsequent changes in the dentition until the end of puberty. In contrast, cysts of inflammatory origin such as the radicular cysts, are much less frequently found in children and more often in adults as a result of pulp necrosis or insufficient root canal therapies.[5,10] More specifically, Shear,[5] in his review of 2,616 jaw cysts in the general population, found 52.3% radicular or residual cysts, followed by dentigerous cysts (16.6%), primordial or odontogenic keratocysts (11.2%), nasopalatine cysts (11%), and paradental cysts (2.5%). Globulomaxillary traumatic aneurysmal bone cysts, eruption cysts, and mucosal cysts of the antrum were found in less than 1% of his cases. Similar findings were presented by Ochsenius et al.[9] In 2,944 odontogenic cysts in the general population, 50.7% radicular cysts, 18.5% dentigerous cysts, 14.3% keratocysts, and 11.1% residual cysts were observed.[9] Even in those studies concerning only child and adolescent populations, where radicular inflammatory cysts were found as predominant,[22] the greater proportion was found in adolescents, a finding in accordance with the previously mentioned studies.

Noninflammatory cystic lesions of developmental origin were the predominant cystic type in the present study, as has been shown before in similar studies on child populations.[8,9] The majority of jaw cysts (68.1%) were developmental in origin (dentigerous cysts, eruption cysts, keratocysts, aneurysmal median mandibular

Table I. Data of patients with cystic lesions ordered according to lesion type

Patient no.	*Gender*	*Age (yrs)*	*Type of cystic lesion*	*Location*	*Treatment*
1	M	13	aneurysmal bone cyst	L	removal of content + curettage
4	M	7	aneurysmal bone cyst	L	removal of content + curettage
14	M	10	aneurysmal bone cyst	L	removal of content + curettage
43	M	7	BBC	L	enucleation
10	M	9	BBC	L	enucleation + extraction of #36
19*	M	8	BBC	L	enucleation + extraction of #46
22	M	8	BBC	L	enucleation + extraction of #46
3	F	6	dentigerous	L	enucleation
7	M	11	dentigerous	U	enucleation
9	M	7	dentigerous	U	enucleation
15	F	9	dentigerous	L	enucleation
16	F	15	dentigerous	L	enucleation
18	M	10	dentigerous	L	enucleation
20	M	12	dentigerous	U	enucleation
25	M	10	dentigerous	U	enucleation
26	F	14	dentigerous	U	enucleation
27	F	11	dentigerous	L	enucleation
29*	F	12	dentigerous	U	enucleation
29*	F	12	dentigerous	L	enucleation
29*	F	12	dentigerous	L	enucleation
30*	M	12	dentigerous	L	enucleation
30*	M	14	dentigerous	L	enucleation (different site than before)
33	M	7	dentigerous		enucleation
34	F	12	dentigerous	L	enucleation
35	M	11	dentigerous	L	marsupialization
8	M	6	dentigerous	L	marsupialization + enucleation (2-stage procedure)
39	M	8	dentigerous + mesiodens	U	enucleation
2	M	8	eruption	U	enucleation
21	F	5	eruption	L	enucleation
5	M	2	eruption	U	marsupialization
12	M	12	eruption	U	marsupialization
40	M	10	eruption	L	marsupialization
24	M	7	midline	L	enucleation
13	F	11	mucosal cysts of the maxillary antrum	U	enucleation
32	M	13	mucosal cysts of the maxillary antrum	U	enucleation
42	F	13	nasopalatine	U	enucleation
23	F	13	odontogenic keratocyst	U	enucleation
31	F	10	odontogenic keratocyst	L	enucleation
41	M	10	odontogenic keratocyst	L	enucleation
11*	F	12	odontogenic keratocyst	L	marsupialization + enucleation (2 stage procedure)
11*	F	15	odontogenic keratocyst		enucleation (recurrence)
36	M	13	odontogenic keratocyst	L	enucleation + apicectomy
6	F	7	radicular	L	enucleation
28	F	13	radicular	L	enucleation
19*	F	10	radicular	U	enucleation + apicectomy
37	F	11	radicular	U	enucleation + apicectomy
38	M	11	radicular	U	enucleation + apicectomy
17	M	10	traumatic	L	curettage

Patient no. indicates the presenting order of the individual in the department. *L*, Lower jaw, mandible; *U*, upper jaw, maxilla; *BBC*, buccal bifurcation cyst.
*Patient had <1 lesion or operation.

cysts, nasopalatine cysts), the remaining 23.4% being inflammatory (radicular cysts, buccal bifurcation cysts, antral mucoceles) and 8.5% nonepithelial (aneurysmal traumatic cysts). Bodner[8] found a percentage of 70.0% for developmental and 13.3% for inflammatory jaw cysts in children, the remaining being nonodontogenic, and Ochsenius at al[9] found a percentage of 67.6% for developmental and 32.4% for inflammatory odontogenic jaw cysts in children. Regarding keratocysts, which appear to be the cystic lesions most liable to recurrence,[23] the percentage in the present study was 10.6%, similar to the results of Ochsenius et al[9] (14.3%

Fig. 5. Orthopantomogram (OPG) of a 12-year-old girl showing 3 dentigerous cysts: 2 in the mandible and 1 in the maxilla. (Courtesy of Dr. P. Christopoulos, Lexturer of Oral and Maxillofacial Surgery.)

of odontogenic cysts in children) and Shear[5] (11.2% of all cystic lesions of the jaws).

When treating children, factors such as the anatomic differences of the growing facial bones, the existence of tooth germs in the jaws, the faster bone repair process compared with adults, and the difficulties in cooperation have to be taken into consideration. For this reason, general anesthesia, inevitable in large-sized cysts or in difficult-to-approach sites, was used in all of our patients, enabling the surgical team to work safely and effectively.

In the present study, the treatment of choice for intraosseous cystic lesions was their surgical removal, aiming to the removal of the cystic lesion in a single-stage procedure, with preservation of the involved permanent teeth and surrounding tissues; this is crucial, because many structures in this age group have not reached their full development. The way to achieve this result was by enucleation in the majority of the lesions (74.5%) and by marsupialization to a less extent (17.0%). Enucleation was meticulous regarding the dental germs and adjacent vital structures, resulting usually in uneventful healing and fewer visits than in case of marsupialization. This treatment option was in contrast to the approach of Bodner et al.,[24] who performed marsupialization in the majority of cases (43%) and less enucleation (35%).

Considering the fact that the pediatric group of patients is characterized by rapid healing and remodeling potential resulting from the excellent blood supply and the ongoing growth procces,[25] autologous bone grafting was not considered to be necessary, even in cases of extended cystic lesions. Postoperative radiographs of our patients justified this choice. This surgical approach was again in contrast with Bodner,[8] who performed enucleation with bone grafting in 22% of cases.

A strict follow-up schedule, every 3 months initially, every 6 months for 2 years, and annually for another 2-3 years depending on the type of the cystic lesion, proved to be very effective, not only in cases of odontogenic keratocysts where recurrence occurs as high as in 60% of the cases,[3] but also in other cases where new cysts developed in different sites (patients no. 19 and 30).

Special care was given to maintain all involved anterior permanent teeth inside or adjacent to the cystic lesions. These were finally erupted either spontaneously or after orthodontic treatment. In 3 of the buccal bifurcation cyst cases, the involved first permanent mandibular molars were extracted, although a more conservative approach has been recommended.[26,27] These patients in the present study were referred for orthodontic treatment aiming to space closure.

It is worth mentioning that although in our material no cases of odontogenic cysts of developmental origin presented malignant degeneration, such a finding has been reported previously.[28,29]

In conclusion, in the present study intraosseous cystic lesions in children were found to be developmental in origin in 68.1% of the cases. Treatment of choice was enucleation in 74.5% of the cases, and involved teeth were preserved in the anterior jaw region and their eruption monitored accordingly. No bone grafting was used when cysts were operated. Long follow-up period allowed early diagnosis of occasional recurrence. Although cystic lesions are benign lesions, in children they may present clinical problems, namely, rapid increase in size, occasional recurrence (odontogenic keratocysts), or severe hemorrhage (aneurysmal cysts). Because dentigerous cysts are by far the most frequent cysts in children, careful clinical and radiologic examination in cases of missing teeth, eruption delay, and swelling of the alveolar process are recommended.

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