

A retrospective analysis of the characteristics, treatment and follow-up of 26 odontomas in Greek children

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Abstract: Odontomas represent the most common type of odontogenic jaw tumors among patients younger than 20 years of age. Clinically, they are often associated with eruption failure of adjacent permanent teeth, and are classified as compound and complex. The aim of the present retrospective study was to present the characteristics, treatment approach and outcome of odontomas in Greek children, over a ten-year period. Twenty six patients, 2 to 14 years of age (mean 9.3 years), with odontomas treated during the years 1999-2008 at the Department of Oral & Maxillofacial Surgery of a Children's Hospital, were included in the study. Data from patients' files were retrieved and they were recalled for review. Odontomas were equally distributed in the maxilla and mandible and 42.3% of them were located in the anterior maxilla. Of the odontomas, 80.7% were related to disturbances in tooth eruption. Bone expansion was observed in 65.3% of the cases. All odontomas were surgically removed, and related impacted permanent teeth were either left to erupt spontaneously, orthodontically guided into occlusion or were removed. Orthodontic intervention appeared to be necessary in older children, while in younger children spontaneous eruption was frequent. In the present study, odontomas were associated with unerupted or impacted teeth. Radiographic examination was essential to verify the presence of the tumor and early removal prevented tooth eruption failure and disturbances in a majority of the cases. (*J Oral Sci* 52, 439-447, 2010)

Keywords: odontogenic tumors; odontomas; children; surgery.

Introduction

Odontomas represent the most common type of odontogenic jaw tumors among patients younger than 20 years of age (1-3). These tumors have also been considered as tumor-like malformations or hamartomas of dental tissues or as developmental anomalies rather than true odontogenic neoplasms (4-6). Their pathogenesis is associated with trauma during primary dentition, hereditary anomalies (Gardner's syndrome, Hermann's syndrome, basal cell nevus syndrome), odontoblastic hyperactivity or alterations of the genetic components responsible for controlling dental development (2,7-10).

Two main, distinct types are acknowledged histologically, while co-existence of both types has occasionally been reported (7,11). Compound odontomas are malformations involving all normal dental tissues and have numerous tooth-like structures (with altered size and shape) known as denticles (12). The other type, known as complex odontomas, are well formed tumors with dental tissues in disorganized distribution (5,13,14).

Odontomas are mainly intraosseous lesions, although location in gingival soft tissues has been reported (15), and are most commonly found in the permanent dentition and less in association with primary teeth (8,16). Anterior maxilla, followed by anterior mandible and postero-inferior regions are the most common locations (7,13).

Clinically, odontomas are frequently associated with eruption failure of permanent teeth, often with delayed exfoliation of primary teeth (4,5); symptomless swelling of the jaw or mild pain on palpation may be present, depending on the size, the type and the location. Being

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usually asymptomatic, odontomas may be diagnosed accidentally in routine radiographs, presenting as well circumscribed tumors with radiopaque and radiolucent areas resembling a tooth, depending on the type and the degree of calcification (7).

Although the diagnosis of odontomas in most cases can be provisionally confirmed by radiographic examination, differential diagnosis includes other odontogenic tumors such as ameloblastic fibroma and fibroodontoma, as well as odontoameloblastoma (14).

Surgical removal is the treatment of choice for odontomas. Care should be taken, however, not to harm adjacent permanent teeth and germs in children, while follow-up is essential for evaluation of further development of the permanent dentition at the removal location (5,14).

Since odontomas are rather common tumors, several papers from different countries can be found in the literature, either as case reports or as retrospective studies. It appears though that there is insufficient data on children from south-eastern Europe.

The aim of this retrospective study was to present the characteristics as well as the treatment and outcome of odontomas in Greek children up to 14 years of age, during a ten-year period.

Materials and Methods

All young patients with odontomas treated in our

Department from January 1999 to December 2008 were included in the study. Data from the patients' files were retrieved, including age, gender, clinical and radiographic appearance, size and location of the tumor, treatment, outcome and follow-up. Regarding tumor location, odontomas were registered as anterior when in the incisor and canine area, middle zone when in premolar area and posterior when in molar area. Patients were then recalled for the study and their condition at the time was recorded (Table 1).

Patients had been referred from general dental practitioners, pediatric dentists, orthodontists or pediatricians, mainly due to disturbances in tooth eruption as well as for bone swelling. Diagnosis of odontomas was based on clinical and radiographic findings; impaction of deciduous or permanent teeth was verified in panoramic, periapical and occlusal X-rays or even CT and CBCT, whenever required. All tumors were surgically removed, with an intraoral approach, under local or general anesthesia. Adjacent deciduous teeth were removed and permanent teeth and germs were preserved whenever not involved by the tumor. Impacted permanent teeth were either left to erupt spontaneously or orthodontic traction was initiated either at the same or at a later stage in collaboration with orthodontists. Few of these teeth, posterior mainly, were removed. Retrieved tissues were examined histologically. Young patients were regularly recalled for clinical and

Table 1 Analytic details of the 26 cases of odontomas in Greek children

Patient no.	Age (yrs)	Gender	Location	Clinical/radiographic findings	Diagnosis	Treatment	Follow-up
1	13	?	UR, anterior	Buccal intraoral swelling, 12 impacted	Complex	Tumor removal, 12 bracket	12 erupted with orthodontic traction
2	9	?	UL, anterior	Buccal intraoral swelling, 21, 22 impacted	Compound	Tumor removal	21, 22 erupted spontaneously
3	14	M	L, anterior middle line	no teeth involvement	Compound	Tumor removal, no teeth involvement	No teeth involvement
4	14	M	UL, posterior	Intraoral swelling, 26, 27 impacted and involved in tumor, sinus involvement	Complex	Tumor and 26,27 removed	No further treatment
5	12	M	L, bilateral posterior	Buccal intraoral swelling, 37, 47 impacted, involved in tumor	Complex	Tumors and 37,47 removed	No further treatment
6	4	M	LR, anterior	Buccal intraoral swelling	Complex	Tumor and 61, 62 removed	Incisors erupted spontaneously
7	8	?	LR, middle zone	Buccal intraoral swelling	Complex	Tumor and 75 removed	Premolars erupted spontaneously
8	9	?	UL, anterior	52 retained, 12 impacted	Compound	Tumor and 52 removed, 12 bracket	12 erupted with orthodontic traction
9	4	?	LL, posterior	Buccal intraoral swelling, 75 impacted, 35 bud involved in tumor	Compound	Tumor and 75, 35 bud removed	Under follow-up
10	13	M	UR, anterior	52 retained, 12 impacted	Compound	Tumor and 52 removed, 12 bracket	12 erupted with orthodontic traction
11	8	?	UR, anterior	Buccal intraoral swelling, 51 retained, 11 impacted	Compound	Tumor and 51 removed	11 bracket 2 yrs post-op, under follow-up
12	13	?	LL, anterior	73 retained, 33 impacted	Complex	Tumor and 73 removed, 33 bracket	33 in improved position, still not in the mouth
13	2.5	?	UR, anterior	Buccal extra oral swelling, 53 impacted	Compound	Tumor and 53 removed	Under follow-up
14	4	M	UR, middle zone	Buccal intraoral swelling	Compound	Tumor and 54 removed	Under follow-up
15	11	?	UR, anterior	Buccal intraoral swelling, 51,52 retained, 11 impacted	Complex	Tumor and 51, 52 removed, 11 bracket	11 in improved position, still not in the mouth
16	11	?	LR, middle zone	Buccal intraoral swelling, 44 malpositioned and involved in tumor	Compound	Tumor and 44 removed	Under orthodontic treatment
17	8	M	UL, anterior	61 retained, 21 impacted	Compound	Tumor and 61 removed, 21 bracket	21 erupted with orthodontic traction
18	9	M	LR, posterior	47 bud, malpositioned	Compound	Tumor removal	No further information
19	2	M	LL, posterior	Buccal fistula, 74,75 impacted	Complex	Tumor removal, 74,75 removed	Under follow-up
20	8	M	UR, anterior	Buccal intraoral swelling	Compound	Tumor and 63 removed	Under follow-up
21	8	M	LR, middle zone	Buccal intraoral swelling, 85, 45 impacted	Compound	Tumor and 85, 45 removed	Under orthodontic treatment
22	11	?	LR, anterior	Buccal intraoral swelling, 43 impacted, severely malpositioned	Complex	Tumor and 43 removed	Closure of the space orthodontically
23	12	?	UR, anterior, palatally	63 retained, 23 impacted	Complex	Tumor and 63 removed	Under follow-up, 23 in improved position,
24	8	M	UL, anterior	Buccal intraoral swelling, 11 impacted	Compound	Tumor removal, 11 bracket	Under follow-up
25	13	M	LR, posterior	Buccal intraoral swelling, 47 impacted	Compound	Tumor and 47 removed	No further treatment
26	14	M	LR, anterior	Buccal intraoral swelling, 42, 43 impacted	Complex	Tumor removal, 42, 43 bracket	Under follow-up

radiographic examination for at least 3 years postoperatively.

Results

Age and gender

Twenty six patients, 14 boys and 12 girls, 2 to 14 years of age (mean 9.3 years) were treated, during the 10-year period (Table 1).

Clinical findings

No history of pain was recorded in the presenting symptoms. The main presenting sign was the eruption failure of permanent teeth. Twenty eight teeth in 21 cases (80.7%) were affected by odontomas, 23 permanent and 5 primary. In 7 of the cases of permanent teeth impaction

(7/23, 30.4%), 8 predecessor primary teeth had failed to exfoliate on time (Fig. 1). Bone expansion was observed intraorally in 17 cases (65.3%), whereas extraoral swelling of the maxilla was registered in only 1 case (Table 1). Fistula formation was the initial symptom in 1 case of odontoma of the primary dentition. In 7 cases (7/26, 26.9%), odontomas were diagnosed incidentally before the expected eruption of the adjacent permanent tooth when radiographic investigation of an intraoral swelling was performed; in these cases, tumors were removed without any interference with local dental development.

Radiographic findings

The precise location, the size and the relation of odontomas to adjacent tissues, were estimated on radiographs. All odontomas were intraosseous, associated with the primary dentition in 5 cases (19.2%), the remaining 21 (80.8%) cases being with the mixed or permanent dentition. A radiopaque mass was most commonly surrounded by radiolucent areas (Fig. 2). Odontomas were equally distributed in the maxilla and mandible (13 cases in each, 50.0%). Anterior maxilla was by far the most frequently involved region (11 cases, 42.3%), followed by equally distributed cases in the anterior and posterior mandible and posterior maxilla (5 cases in each region).

In most of the cases, odontomas were approximately up to 1 cm in diameter, but in 6 cases their diameter was larger, ranging from 1.5 to 3 cm. Consequences of the odontomas on the adjacent dentition were registered; 5 primary teeth, 1 canine and 4 molars, were found impacted. Permanent teeth were impacted and/or displaced as follows: 23 teeth in 19 (73.0%) patients; 12 in the maxilla in 10 patients (9 incisors, 1 canine and 2 molars) and 11 in the mandible in 9 patients (1 incisor, 3 canines, 3 premolars and 4 molars) (Fig. 3 and Table 1). In one case of maxillary odontoma, the tumor penetrated the maxillary sinus and

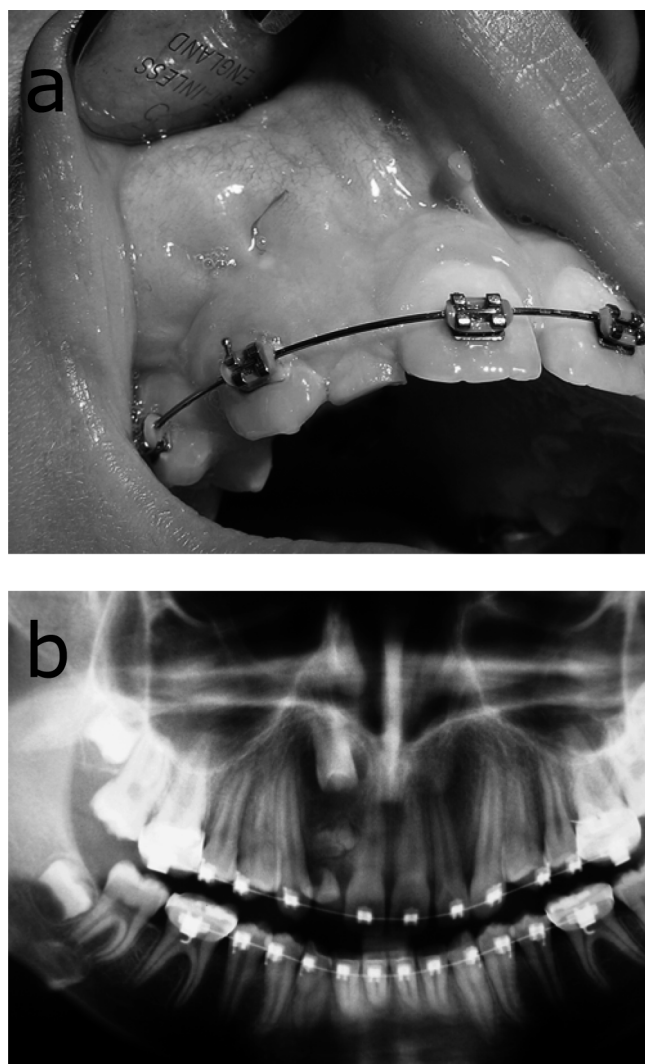


Fig. 1 Odontoma in anterior maxilla. a) clinical appearance with the primary lateral incisor retained. b) radiographic appearance: odontoma obstructed the eruption of permanent lateral incisor.

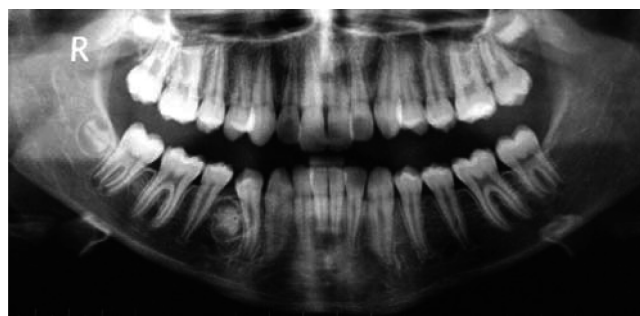


Fig. 2 Radiographic appearance of odontoma at the lower right middle zone. Note the malpositioned first premolar involved in the tumor.

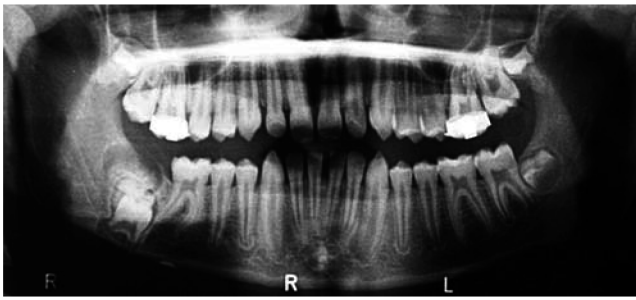


Fig. 3 Odontoma in lower posterior mandible which obstructed the eruption of second permanent molar.

the first and second permanent molars were displaced cephalade. In one interesting case of mandibular odontoma, the location was close to the chin and did not obstruct the dentition development.

Treatment

All odontomas were removed either under local or general anesthesia through an intraoral approach. Intraoperatively, the buccal cortex was found to be thin or perforated in 17 cases (65.3%). All 5 impacted and 8 retained primary teeth were removed. Fourteen of the impacted 23 permanent teeth (60.9%, 10 incisors, 3 canines, 1 molar) were maintained after tumor removal and 9 were removed (39.1%, 1 canine, 3 premolars, 5 molars) (Table 1). Four out of the 14 impacted teeth (28.6%) were left to erupt spontaneously after odontoma removal and the remaining 10 (71.4%) had an orthodontic device for traction attached either during the operation or at a later stage (Fig. 4). Post operative healing was uneventful in all cases.

Pathological diagnosis

Dental structures at different developmental stages and odontogenic epithelium were found. Odontomas were compound in 15 cases (57.7%) and complex in 11 cases (42.3%) (Fig. 5); a maxillary predilection of compound odontomas (9/15), and a mandibular predilection of complex (7/11) were identified.

Follow-up and outcome

All young patients were regularly examined for at least 3 years postoperatively and at the time of the final recall for the purpose of the study. Bone healing was verified radiographically in all cases with no recurrence, and continuous dental development was recorded in a majority of the cases. When odontomas were associated with primary dentition and in most cases of early mixed dentition

(10 cases in all, patient nos. 2, 6, 7, 9, 13, 14, 18, 19, 20, 23, Table 1), no orthodontic intervention was made. Four of the above mentioned very young children (patient nos. 9, 13, 14, 19) and 2 older ones (patient nos. 20, 23) are still under follow-up presenting normal dentition development. In 3 more cases (patient nos. 2, 6, 7), anterior teeth erupted spontaneously and in 1 case (patient no. 18) contact with the patient was lost and further information could not be obtained. In cases where orthodontic traction was exercised, 4 impacted teeth were in occlusion (patient nos. 1, 8, 10, 17), 3 were still under treatment but not yet in position in the mouth (patient nos. 11, 12, 15), and 2 of the more recent cases of impacted teeth are under follow-up (patient nos. 24, 26). No noteworthy difficulty was registered in cases in which permanent teeth were removed. In 3 cases of premolars and the sole canine removed (Fig. 6), spaces in the dentition were closed orthodontically (patient nos. 16, 21, 22) and in those cases where molars were removed, there was no further treatment (patient nos. 4, 5, 25). In one case, no teeth were involved (patient no. 3).

Discussion

Etiology

Although odontomas have been associated occasionally with hereditary anomalies, no syndromes were evident in any of our cases. In 3 of the cases of odontomas of the anterior maxilla, previous trauma at the site had been reported. Trauma has been implicated as a possible etiological factor for odontoma formation, but no documented data appear in the literature (7,8,14).

Frequency

In agreement with previous studies (6,7), odontomas were the most frequent odontogenic tumors treated in our department (26/40, 65.0%), over the 10-year period. In a review of studies on odontogenic tumors, it was found that odontomas were the most frequent ones in many countries such as the USA, Canada and Finland. On the contrary, in other countries (India, China, African countries), the incidence of odontomas was much lower and these variations have been attributed to geographic, ethnic, and socioeconomic factors (6,17-19).

Age and gender

Studies on odontomas include patients of all ages (14). Although there are several case reports of odontomas in children (2), not many studies include only children. According to the literature, odontomas occur most frequently in the second decade of life (1,4,6,7). The mean age of patients (9.3 years) in the present study showed a

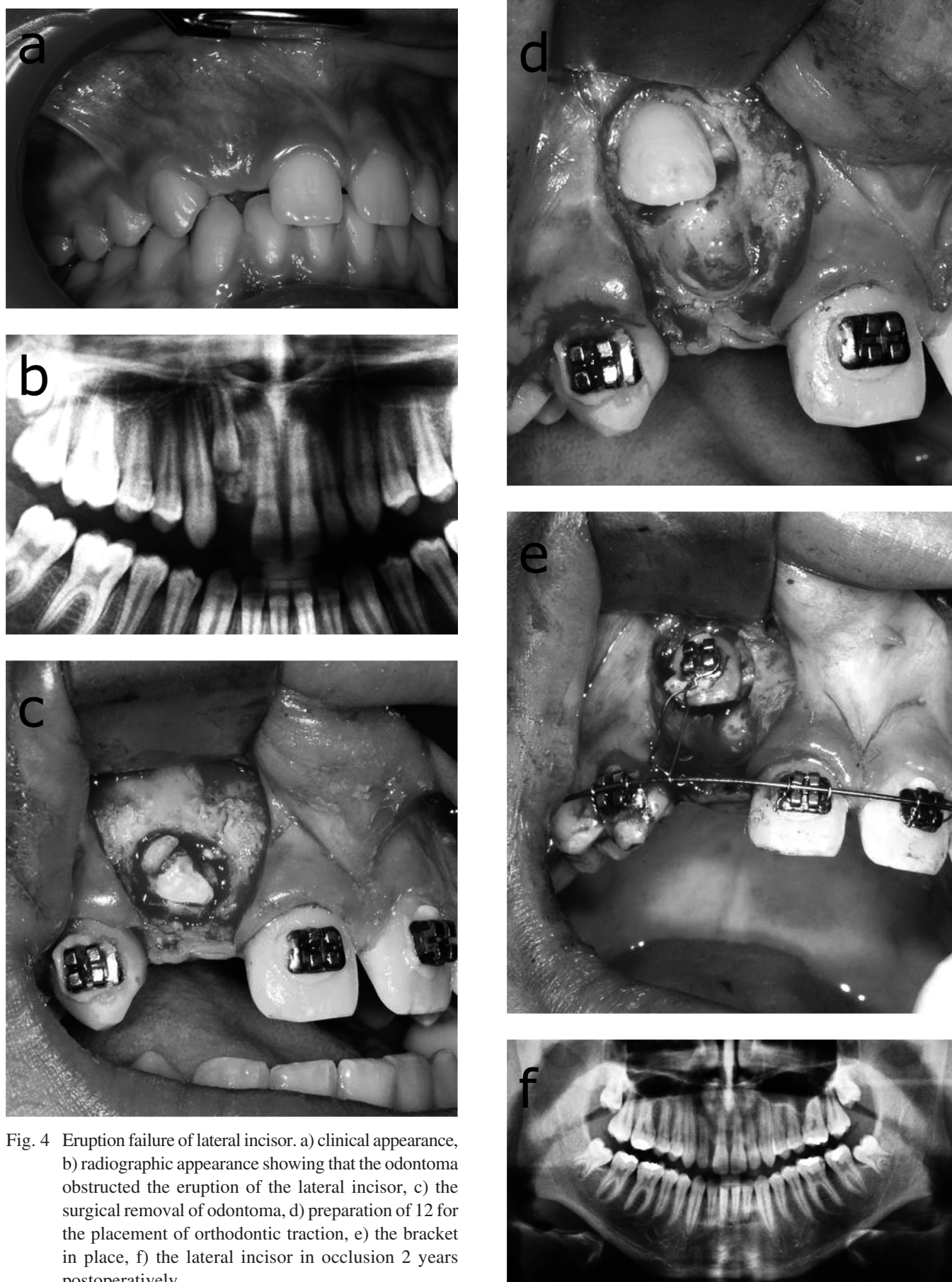


Fig. 4 Eruption failure of lateral incisor. a) clinical appearance, b) radiographic appearance showing that the odontoma obstructed the eruption of the lateral incisor, c) the surgical removal of odontoma, d) preparation of 12 for the placement of orthodontic traction, e) the bracket in place, f) the lateral incisor in occlusion 2 years postoperatively.

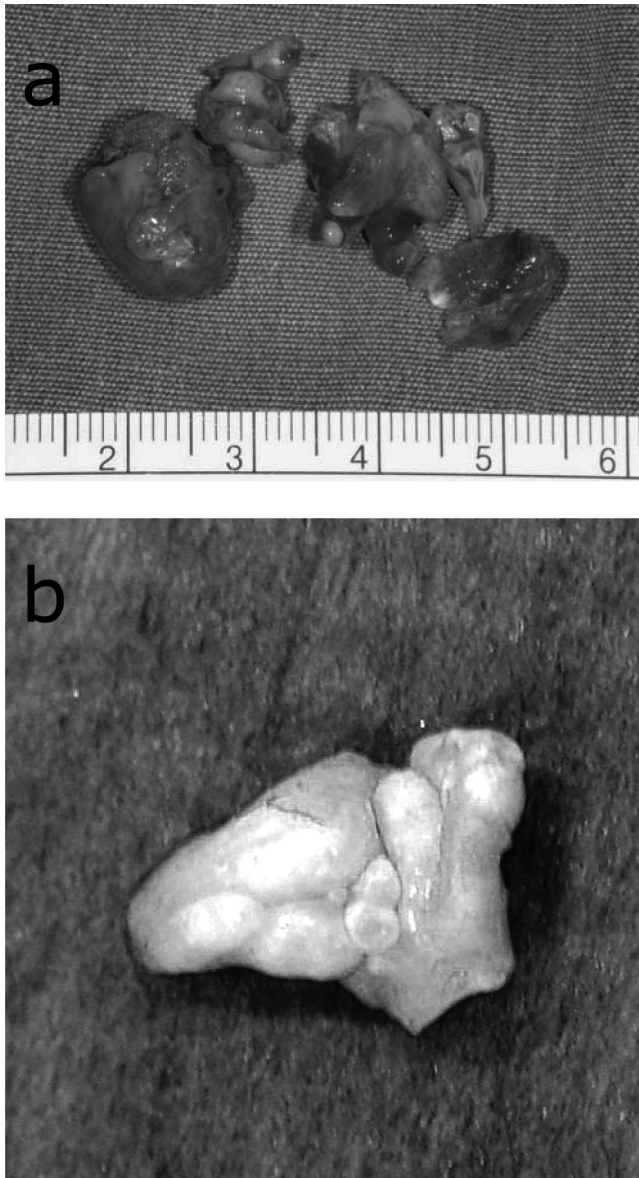


Fig. 5 Gross appearance of odontomas. a) Complex odontoma of case shown in Fig. 3, b) Compound odontoma of case shown in Fig. 4.

tendency towards the second decade. Additionally, the finding of this study that 53.8% of the patients were under 10 years old was in agreement with the 50% previously reported in children (5).

Findings of the study regarding the rarity of odontomas in the primary dentition (5/26, 19.2%), were higher from the previously reported 2% and 14.7% (1,5). Contemporary radiographic techniques following parental and professional alert regarding dentition disturbances may be the reason for such an early diagnosis of odontomas.

In the present study, in agreement with previous findings (7,20,21), a male predilection of 53.8% was found. In

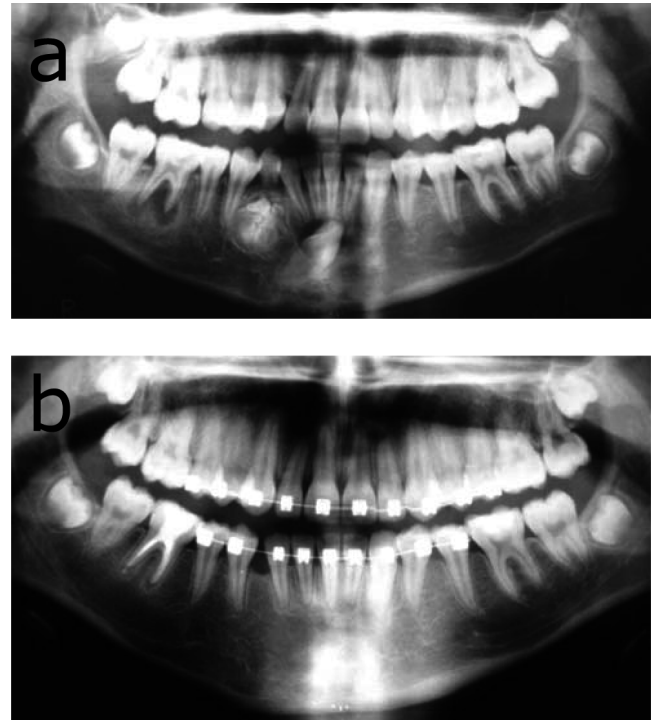


Fig. 6 Odontoma in anterior mandible. a) primary canine retained in occlusion and 43 impacted and malpositioned. b) odontoma and permanent canine were removed (the canine following orthodontic instructions) and the gap was closed orthodontically.

other studies, on the contrary, no predilection by gender was reported (6,14).

Clinical findings

Intraoral bone expansion with normal overlying tissues, found in 65.3% of the cases, was a parameter not usually recorded by other studies. Extraoral swelling, representing 3.8%, was rather similar to the previously reported 2.5% (5). Additionally, the most common presenting sign, the impaction of permanent teeth, found in 80.7% of our cases, was in agreement with previous studies (5,14). Percentages of tooth eruption disturbances, in studies where patients of all ages were included (1,7,22) present differences ranging from 41.0% to 73.7%. These percentages increased to above 80% when only the children were included (5).

Radiographic findings

Appearance of odontomas was as described and expected. CTs were of assistance whenever in doubt for the exact relation to adjacent teeth.

Location

In all our cases, odontomas were intraosseous and associated in 21 cases (80.7%) with the crown of an impacted tooth; no erupted odontoma was found as previously reported (23). Our findings regarding tumor location, were similar to those of previous reports (7,22,24) which found a rather equal distribution in the jaws, and different from other studies where strong predilection for the maxilla was reported (5,6,15,25,26).

Anterior maxilla was the most common location in the present study, in agreement with findings of other studies on children and adults (5,6). There is no obvious explanation for anterior maxillary predilection of odontomas, although it could be assumed that the subsequent aesthetic impairment leads to early diagnosis and treatment. The size of odontomas in this study did not exceed 3 cm which is in agreement with previous findings (5). The rather small size may be attributed to early diagnosis, as odontomas show similar development to normal dental tissues, increasing therefore progressively in size with advancing age (27). In most cases, their size was close to the size of the permanent tooth affected, without major consequences to other adjacent teeth. Nevertheless, there were some cases of larger-sized tumors obviously influencing more adjacent teeth. In one case, two bilateral odontomas adjacent to lower second molars were removed, in agreement with findings of others who reported cases of large-sized and multiple odontomas (28).

Treatment

In agreement with others (4,6,7,14), surgical removal of odontomas was our treatment of choice. Being, in most cases, well circumscribed tumors, odontomas could be easily excised when surgically exposed. Care was taken not to harm adjacent structures such as root apices and germs. Primary teeth related to the tumor were removed as well, whilst impacted permanent teeth were treated according to their location, their developmental stage and the age of the child. Effort to maintain all front teeth was successful and only one lower canine was removed due to its severe malposition and following orthodontic advice. Teeth were left to erupt spontaneously in younger ages when eruptive ability still exists. Percentages of spontaneous eruption of impacted teeth in such cases has been reported to range from 32% to 48.5% (5,29). In cooperation with orthodontists and in agreement with previous studies (30,31), traction was applied whenever the apex of the tooth was closed or almost closed. Posterior teeth impacted due to odontomas were removed together with the tumor when severely malpositioned without possibility to erupt.

Application of an orthodontic traction device to the

impacted permanent teeth intra-operatively or at a later stage, is a matter of discussion among authors (5,10,31). A waiting period of 3 months for the spontaneous eruption has been proposed (5) whilst others (31) claim that orthodontic therapy is not usually required, in cases of small-sized odontomas (smaller than the impacted permanent tooth). However, in order to avoid a second stage operation, application of an orthodontic device would be advisable during the tumor removal operation, as was the case in all but one of our patients.

Pathology

A majority of the odontomas in this study were compound (15/26, 57.6%) in agreement with literature reports (14), although a higher proportion of 2:1 between compound and complex has been reported (23). The most common location of compound odontomas was found to be the anterior portion of the maxilla (6/15, 40%), in agreement with others (5,7,23); whereas, a predilection for complex odontomas to occur in the mandible (14) was also verified in the present study. Nevertheless, from a clinical perspective, it has been claimed that distinguishing between the 2 types of odontomas is not important (6).

Follow-up

Although the presence of odontomas is indicative of a local developmental disturbance, associated impacted teeth were in most cases affected mechanically only, by the presence of an "obstacle" or "barrier" and therefore their eruption was possible postoperatively. It is essential nowadays to preserve impacted permanent teeth and follow-up their eruption after the removal of odontomas. Findings of the present study showed that obstructed anterior permanent teeth erupted or were expected to erupt in almost all cases, following tumor removal. In very young children, eruption was spontaneous or assisted at a second stage, whereas in children over 8-9 years of age orthodontic traction was applied. In spite of the inadequate numbers for statistical analysis, it appeared that orthodontic traction, whenever required, contributed to successful eruption.

Regarding impacted posterior teeth, the decision to maintain or remove them should depend entirely on the case; in our study a majority of the posterior teeth were heavily involved with the tumor or severely malpositioned and had to be removed.

Odontomas are often associated with disturbances in tooth eruption and impaction of permanent teeth. Apart from the surgical removal of the odontogenic tumor, care should be taken to preserve the disturbed dentition. In children, the impacted permanent teeth, depending on the

age of the child and the tooth development, may be left to erupt spontaneously, or they may be guided to occlusion via orthodontic traction. In any case, follow-up is essential following odontoma excision.

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