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Benign fibro-osseous lesions of the jaws in children. A 12-year retrospective study



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ABSTRACT

Introduction: Fibro-osseous lesions, a diverse group of bone disorders including developmental, reactive or dysplastic diseases and neoplasms, share overlapping clinical, radiographic and histopathologic features and demonstrate a wide range of biological behaviour.

Aim: To evaluate the characteristics, treatment and outcome of benign fibro-osseous lesions of the jaws in children.

Patients and method: All patients with fibro-osseous lesions of the jaws treated at the department of Oral and Maxillofacial Surgery of the «A & P Kyriakou» Children's Hospital of Athens from 2000 to 2011 were included in this study. Data were retrieved from patients' files and their present situation was registered. Results: Sixteen males and 10 females (mean age 8.5 years) were treated. Fibrous dysplasia was most often encountered (26.9%), and the mandible was the most frequent location (76.9%).

All cases were surgically treated and histopathologically confirmed. Marginal ostectomy was performed in 7 cases, partial ostectomy in 4, enucleation and curettage in 10 and trimming-remodelling in 5 cases. Mean follow-up was of 5.5 years with no recurrence, except in one case of fibrous dysplasia. *Conclusions*: Fibro-osseous lesions, although sharing similar microscopic features, exhibit a variety of clinical behaviour rendering their treatment highly individualized.

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1. Introduction

A diverse group of osseous disorders including hereditary or developmental lesions, reactive or dysplastic diseases and neoplasms have been described as benign fibro-osseous lesions (Alawi, 2002; McDonald-Jankowski, 2004; Mehta et al., 2006). They frequently develop in the craniofacial skeleton and especially in the jaws, the nasal cavity, the paranasal sinuses and the orbit and are mainly characterized by replacement of bone by a connective-tissue matrix (Koury et al., 1995; Choi et al., 2000; Brannon and Fowler, 2001; Mehta et al., 2006).

Fibro-osseous lesions (FO lesions) of the maxillofacial bones share overlapping clinical, radiographic and histopathologic features that may lead to diagnostic confusion and difficulty in differentiation (Mafee et al., 2003). There is a relative disagreement among authors in pertinent literature, about their classification and this issue has clinical implications, as the wide range of biologic behaviour these lesions demonstrate requires different management strategies (Papadaki et al., 2005; Mehta et al., 2006).

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Although the term fibro-osseous lesion had not been included in the WHO's classification of 1992 (Kramer et al., 1992), these lesions were formally re-classified in 1993 (Kramer et al., 1993) and have been included ever since, based on their biological behaviour and histopathology and in agreement with Waldron's recommendations of 1985. Thus FO lesions nowadays constitute a group of "neoplasms and other tumours related to bone" (Brannon and Fowler, 2001; Kramer et al., 1993; McDonald-Jankowski, 2004).

The main clinical symptom is soft tissue swelling and enlargement of the affected bones, which may lead to cosmetic and functional disturbances. The presence of pain, paraesthesia, trismus or dental occlusal findings have been reported, depending on location. Alternatively FO lesions may be completely asymptomatic, identified only on routine radiographs (Slootweg et al., 1994; Abdelsayed et al., 2001; Sarwar et al., 2008), while fibrous dysplasia can be associated with generalized endocrinopathy (Alawi, 2002; Troulis et al., 2004).

The radiological appearance of fibro-osseous lesions varies depending on the stage of development. In the early stages the lesion is radiolucent and well-defined, while at later stages it changes into radio-opacity with ill-defined borders. The radiographic appearance may thus be either a radiolucent, a mixed

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radiolucent—radiopaque, a predominantly radiopaque, or ground-glass appearance (Araghi and Haery, 1993; Alawi, 2002). In craniofacial lesions the bone appearance has been sub classified into three different patterns: pagetoid, cystic or sclerotic (Panda et al., 2007).

The treatment of FO lesions, generally based on their biological behaviour and regional aggressiveness, is highly individualized. A conservative approach is indicated in some cases, and a more radical approach including a surgical resection may be warranted in others (Koury et al., 1995; Panda et al., 2007).

Fibro-osseous lesions in adults have been well documented in many studies and although they often occur in children and adolescents, relevant reports are rather infrequent (Khanna et al., 1979; Parham et al., 2004).

The purpose of this retrospective study was to evaluate the characteristics, treatment and outcome of benign fibro-osseous lesions of the jaws, in children and adolescents.

2. Material and Methods

All patients with fibro-osseous lesions of the jaws, treated at the department of Oral and Maxillofacial Surgery of the "A. & P. Kyriakou" Children's Hospital of Athens for a 12-year period (2000—2011) were included in this study. The criteria for inclusion of the various lesions were their prior classification as fibro-osseous lesion in previous studies.

Data from the patients' files including age, sex, location, type of lesion, applied treatment and outcome, were registered. Most patients, were still under follow-up and were recalled for examination.

3. Results

Twenty-six Caucasian patients were included in the study. There was a male predilection with 16 males (61.5%) and 10 females

(38.5%), with a mean age of 8.5 years (range from 4 months to 17 years). The mean age for male patients was 8 years (4 months—17 years), while the mean age for female patients was 9.5 years (6 months—16 years). Fibro-osseous lesions represented the 12.6% of all benign tumours in children treated in our department for the same period of time (latrou et al., 2012).

There were 7 cases of single fibrous dysplasias, one case of McCune—Albright syndrome, one juvenile aggressive ossifying fibroma, 3 central ossifying fibromas, 3 unclassifiable fibro-osseous lesions-dysplasias, one case of osteoblastoma, one desmoplastic fibroma, 5 aneurysmal bone cysts with high presentation of fibrous-osseous elements and 4 cases with myofibromatosis, affecting the mandible.

All parameters and findings registered have been summarized in Table 1.

The most frequent location was the mandible (20 cases, 76.9%); in 15 cases, the mandibular body, in 3 cases the angle of the mandible and in 2 cases the ramus. In 16 cases the lesions originated from the body of the mandible, while in 4 cases of myofibromatosis, the mandible was affected by expansion from the adjacent soft tissues. As far as the maxilla was concerned, the bone was affected in 5 cases of fibrous dysplasia and one case of aneurysmal bone cyst where the whole maxilla was destroyed and the lesion had expanded into the naso—ethmoid complex (Table 1).

The main clinical symptom for all patients was swelling or enlargement of the jaw; in 4 cases (15.4%) pain was reported (4 cases of myofibromatosis) and there was progressive trismus in one case of desmoplastic fibroma. Teeth motility and/or displacement were noticed in few cases, where the dentate part of the jaw was affected.

CT scans and simple X-rays were available in all cases. Radiologic examination showed a radiolucent (cystic) appearance in 15 cases (57.7%), pagetoid (sclerotic) appearance (Fig. 1a and b) in 5 cases (19.2%) and mixed in another 6 cases (23.1%) (Table 1).

Table 1
Data of the patients included in the study, concerning gender, age (in years), type of the lesion, localization, radiographic findings, treatment and time elapsed between surgery and re-examination (in years).

No.	Sex	Age	Type	Localisation	Radiographic appearance	Treatment	Follow-up (years)
1	M	13	ABC	Mandible (body)	Radiolucent	Enucleation + curettage	12
2	M	11	Mc-As	Mandible (body)	Radiolucent	Partial ostectomy	10.5
3	F	7	FM	Mandible (body)	Radiolucent	Marginal ostectomy	9.5
4	M	11	ABC	Mandible (body)	Radiolucent	Marginal ostectomy	9
5	M	14	DF	Mandible (ramus)	Mixed	Enucleation + curettage	8.5
6	M	5	ABC	Mandible (angle)	Radiolucent	Enucleation + curettage ^a	8.5
7	M	2.5	FM	Mandible (1 border)	Radiolucent	Marginal ostectomy	7
8	F	7	COF	Mandible (body)	Mixed	Enucleation + curettage	7
9	M	13	UFOD	Mandible (body)	Radiolucent	Enucleation + curettage	7
10	M	3	UFOD	Mandible (angle)	Mixed	Enucleation + curettage	6
11	F	14	FD	Maxilla (both sides)	Pagetoid (sclerotic)	Trimming + remodelling	5.5
12	M	6	FM	Mandible (1 border)	Radiolucent	Partial ostectomy	5
13	F	5	FD	Maxilla	Pagetoid (sclerotic)	Trimming + remodelling	5
14	F	16	FD	Mandible (body)	Pagetoid (sclerotic)	Trimming + remodelling	4
15	M	3	FM	Mandible (body)	Radiolucent	Marginal ostectomy	4
16	M	17	JOF	Mandible (body)	Mixed	Partial ostectomy ^a	4
17	F	0.5	UFOD	Mandible (ramus)	Radiolucent	Enucleation $+$ curettage	4
18	M	7	FD	Maxilla	Pagetoid (sclerotic)	Trimming + remodelling	3.5
19	F	12	FD	Maxilla	Pagetoid (sclerotic)	Trimming + remodelling	3.5
20	F	14	OB	Mandible (anterior)	Radiolucent	Marginal ostectomy ^a	3
21	M	0.5	COF	Mandible (body)	Radiolucent	Enucleation + curettage	3
22	M	6.5	FD	Maxilla	Mixed	Enucleation $+$ curettage	2
23	F	9	FD	Mandible (body)	Mixed	Marginal ostectomy	1.5
24	F	9	ABC	Ramus of mandible	Radiolucent	Marginal ostectomy	1.5
25	M	11	ABC	Maxilla + nasoethmoid complex	Radiolucent	Partial ostectomy	1.5
26	M	3	COF	Mandible (body)	Radiolucent	Enucleation + curettage	0.5

M: Male, F: Female.

FD: Fibrous dysplasia, **Mc**—**As**: McCune—Albright Syndrome, **COF**: Central ossifying fibroma, **JOF**: Juvenile ossifying fibroma, **UFOD**: Unclassifiable fibro-osseous dysplasia, **OB**: Osteoblastoma, **ABC**: Aneurysmal bone cyst with fibro-osseous elements, **DF**: Desmoplastic fibroma, **FM**: Fibromatosis.

^a Iliac bone grafting.

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