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Radiological Imaging Findings of Craniofacial Fibrous Dysplasia

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ABSTRACT

AIM: To present the radiological findings of fibrous dysplasia (FD) patients who had computed tomography (CT) and/or magnetic resonance imaging (MRI) scans.

MATERIAL and METHODS: This study included a total of 25 patients (17 female and 8 male) who were found to have FD based on CT examinations between March 2010 and July 2018. Involved bones, type of involvement (single or multiple) and CT appearance features (ground-glass, sclerotic, cystic or mixed type) were evaluated.

RESULTS: Age range of the patients with FD was 14-55 (mean: 29.92 ± 12.63) years. Sixteen patients had single bone and nine patients had multiple bones affected. Single bone most frequently involved maxillary bones. Multiple bone involvement affected up to four bones, sphenoid bone being the most frequent. Fifteen lesions were mixed type, eight were ground-glass and two were sclerotic on CT. Intense contrast-enhancement was observed in four patients on MRI.

CONCLUSION: Craniofacial FD is more frequent in young adults and women, and more commonly involves single bone, mostly maxillary and sphenoid bones. Craniofacial FDs could be of different types on radiological examinations depending upon their compositions.

KEYWORDS: Computed tomography, Craniofacial bones, Fibrous dysplasia, Magnetic resonance imaging

INTRODUCTION

Fibrous dysplasia (FD) is a congenital, but not hereditary, non-neoplastic benign bone disease in which structurally weak fibrous and osseous tissue takes the place of normal medullar bone (1-3,8,15,20,23). FD was first described in 1891 by von Recklinghausen, detailed in 1937 by McCune and Brunch and termed fibrous dysplasia by Lichenstein in the following year (2). FD could lead to expansion, weakness and distortion of bone (17). It could be in the forms of monostotic (MFD) involving a single bone or polyostotic (PFD) involving multiple bones (1-3,8,15,17,19,20,23). In addition, PFD could manifest itself as a component of McCune-Albright syndrome characterized by stains on skin and endocrinopathies (2). FD could involve any bone of body, but most frequently it involves proximal bone followed by femur, tibia, humerus, costae and craniofacial bones in decreasing order (2). Cranial or facial involvement is observed in about 25-30% of all FD patients (2,12). Craniofacial involvement affects maxillary and mandibular bones most, while frontal, sphenoid and ethmoid bones are less influenced (12). Occipital and temporal bones, on the other hand, are rarely affected (1,3,12,19,20,23). In the present study, radiological imaging findings of patients who were found to have FD based on computed tomography (CT) and/or magnetic resonance imaging (MRI) performed for craniofacial region due to various reasons were discussed.

MATERIAL and METHODS

The present retrospective study was approved by institutional local ethics committee (No. 18-KAEK-119). A total of 25 patients (17 female and 8 male) who were found to have

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FD out of 23,420 patients who had brain, maxillofacial or paranasal CT examinations were included. FD was diagnosed histopathologically in 9 patients and radiologically in 16 patients. Four patients whose lesions could not be distinguished from other bone lesions radiologically (osteoma, ossified fibroma, osteitis condensans, etc.) and who did not have histopathological examinations were excluded. Ten of the patients found to have FD had had MRI. Follow-up radiological examinations were carried out in seven patients for periods of 25-102 months (mean 55.8 ± 28.0). Involved bones, involvement type (single or multiple) and CT appearance features (ground-glass, sclerotic-homogenous dense, lytic-cystic or mixed, i.e., ground-glass with sclerotic and/or lytic type) were studied. Images of the patients were obtained from archive and communication system of our hospital (Sectra IDS7 PACS, Sweden) and were evaluated by two radiologists with 12 (E.G.) and 6 (M.B.) years of work experience.

RESULTS

Age range of patients found to have craniofacial FD was 14-55 (mean: 29.92 ± 12.63). Incidence of craniofacial FD was 0.1%. In 16 patients (64.0%), one bone was involved while multiple bones were involved in 9 patients (36.0%). Involved bones of 16 patients with single bone involvement were maxillary bone in six patients (Figures 1A-C; 2A, B; 3A-D), parietal bone in five patients (Figure 4A-F), frontal bone in two patients and sphenoid, mandibular and temporal bones in one patient each. Multiple bones of up to four were involved, sphenoid bone being the most common one (Figures 5A-D; 6A-F). In terms of CT patterns, 15 patients had mixed type (nine patients had ground-glass + sclerotic, six patients had ground-glass + sclerotic + lytic) while eight patients had pure ground-glass and two patients had sclerotic patterns. Pure cystic pattern was not observed in any patient. Intense heterogeneous contrast-enhancement was observed in four of the five patients who had contrast-enhanced MRI examination (Figure 3A-D), while a contrasting increasing towards the later phases were observed in the fifth patient (Figure 4A-F). Demographic and clinical features of FD patients were presented in Table I, while their radiological findings were given in Table II.

DISCUSSION

Etiology of FD is not fully understood (12). It constitutes 2.5% of all bone tumors and 5-7% of all benign bone tumors

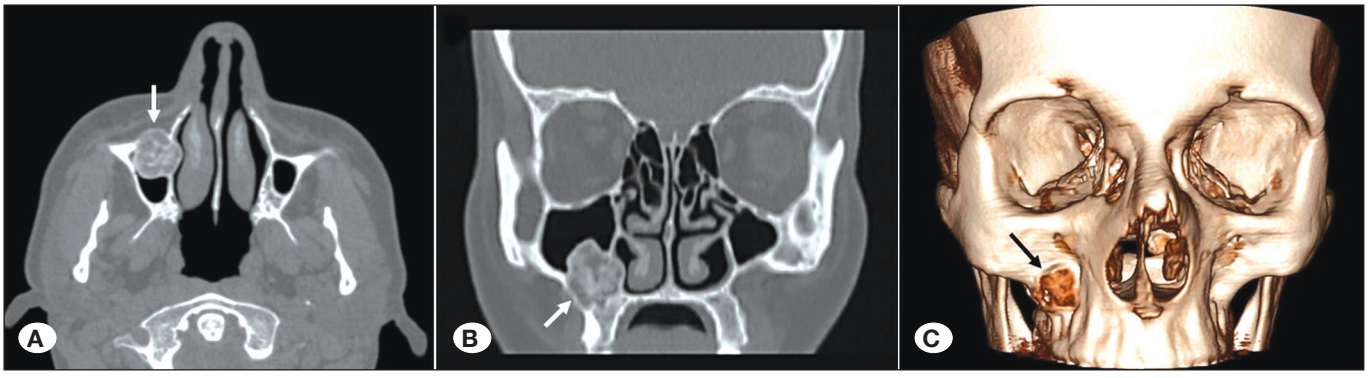

Figure 1: A 35-year-old female patient with palpable swelling in right maxillary region. **A)** Axial, **B)** coronal plane CT slices show a histopathologically verified mixed type FD extending to right maxillary sinus base-medial wall and alveolodental area (write arrows). **C)** Dysmorphic lesion area (black arrow) is shown on right maxillary sinus antero-inferior level on volume rendering CT image.

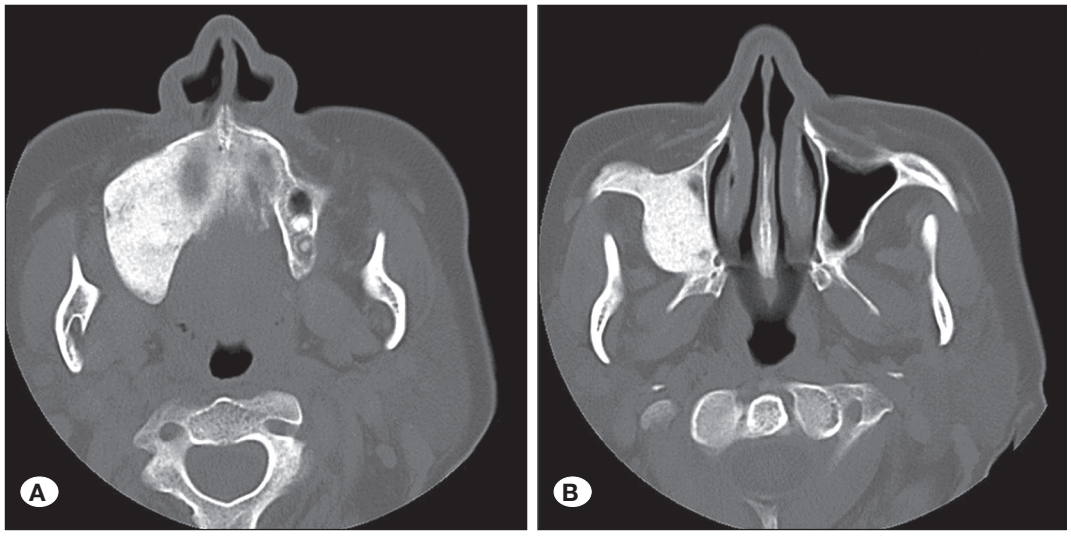

Figure 2: A 48-year-old female patient with stiffness and pain in right upper molar region for three months. **A)** and **B)** Axial plane CT slices show a histopathologically verified sclerosis type FD causing expansion in right maxillary bone.

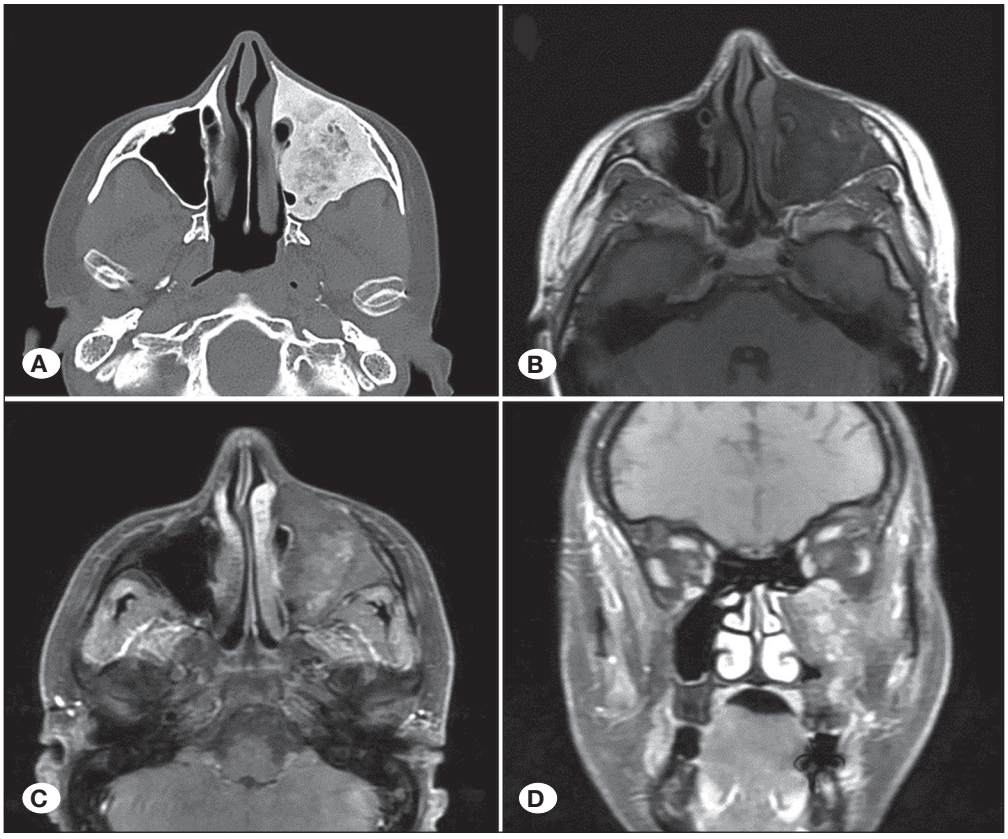

Figure 3: A 31-year-old female patient with facial swelling **A)** Mixed type (ground glass + sclerotic) FD causing a slight expansion in left maxillary bone is shown on CT image. **B)** The lesion appears as slightly heterogeneous and hypointense on T1-weighted axial MR image. **C)** axial and **D)** coronal contrast-enhanced MR images show heterogeneous contrast-enhacement in the lesion.

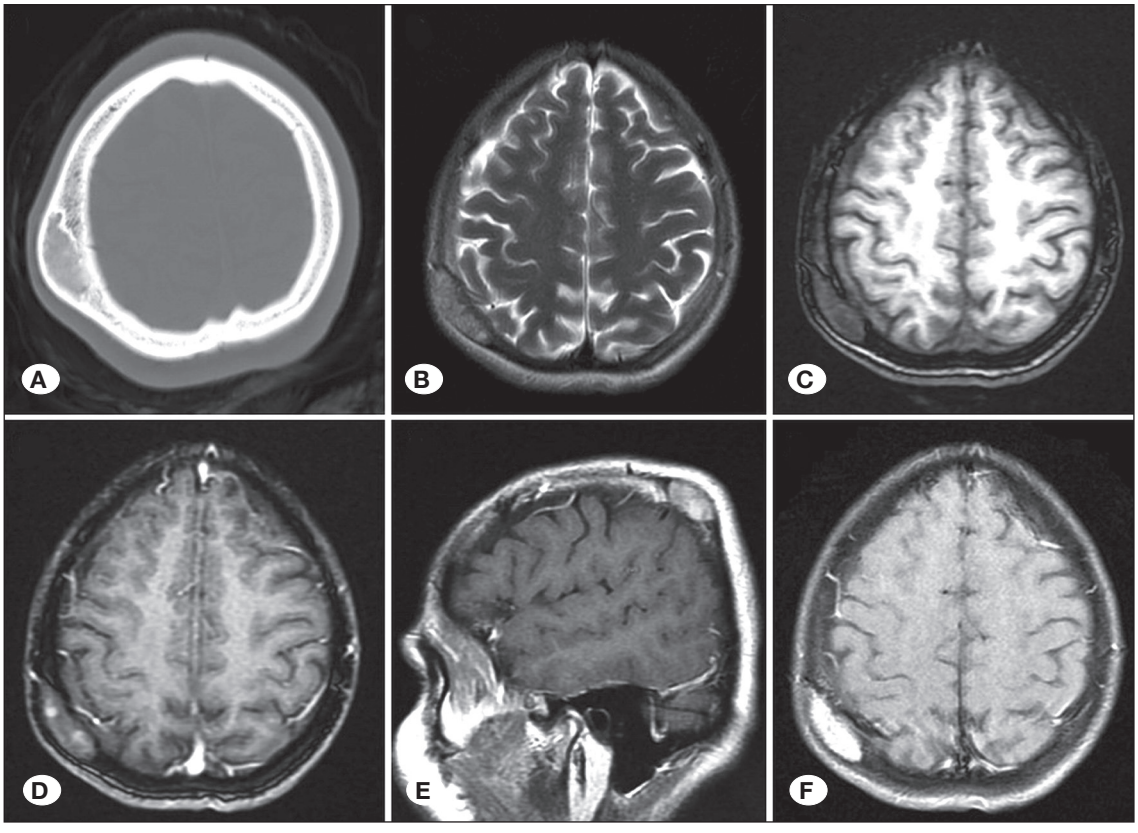

Figure 4: A 27-year-old female patient with swelling on her head since childhood. **A)** Axial CT image shows histopathologically verified fibrous dysplasia in ground-glass density on right parietal bone. Axial plane **B)** T2-weighted and **C)** T1-weighted MR images show slightly hypointense lesion. **D-F)** Focal contrast-enhancement on MR images in early phase **(D)**, increasing contrast-enhancement in later phases **(E)** and homogenous intense contrast-enhancement in late phase **(F)** are shown in axial, sagittal and axial planes, respectively.

(11,12). Its incidence rate in craniofacial bones is not exactly known. In the present study in which CT images of 23,420 patients involving craniofacial areas were studied, radiology-based FD incidence was 0.1%. FD generally affects older age children and young adults (21). Most patients get diagnosis before the age of 30 (12). Atalar et al. studied 32 patients with craniofacial FD and found that mean age of the patients were 28.3 ± 15.3 (2). In the present study, mean age of the patients were 29.92 ± 12.63, being slightly higher than what was reported in literature. In a study carried out by Hanifi et al. on 12 patients with craniofacial FD, only two patients over 30 years of age had diagnosis (12). In the present study, on the other hand, about half of the patients (44.0%) had diagnosis after 30 years of age.

Incidence rates of FD were reported to be similar in males and females. In the study by Hanifi et al. (12) 66.7% of cases were female and 33.3% male, while Atalar et al. reported that 59.4% of cases were female and 40.6 male (2). In the present study, 68.0% of cases were female and 32.0% male, which indicated somewhat higher incidence of cranial FD in females compared to literature. Atalar et al. found similar incidence rates of craniofacial FD involving single bone for females and males, but incidence rate was twice higher in females when multiple bones were involved (2). In the present study, multiple bone involvement was more frequently (66.6%) in women, similar to study by Atalar et al. (2). On the other hand, single bone involvement in this study was about 2.5 times higher in females.

Table I: Demographic and Clinical Findings of Patients with Fibrous Dysplasia

Patient No	Gender	Age (years)	Clinical Findings	Operation	Follow-up time (months)
1	F	35	Left visual loss and trigeminal neuralgia	-	-
2	F	14	Right hard swelling on inferior orbital rim	-	-
3	F	31	Left facial swelling	-	-
4	F	23	Right frontal swelling and visual loss	Partial resection	-
5	M	19	Left parietal swelling	-	-
6	F	55	Left maxillary swelling	Biopsy	-
7	F	26	Left maxillary swelling	-	36
8	F	49	Chronic otitis media	-	-
9	F	40	Right parietal swelling	-	-
10	F	48	Right molar teeth and hard palate swelling	Biopsy	102
11	F	49	Incidental (Right hearing loss)	-	62
12	F	32	Left mandibular swelling	Partial resection	-
13	M	20	Headache and nasal breathing problems	-	27
14	M	15	Right external acoustic channel narrowing and swelling	-	-
15	M	25	Incidental (Trauma)	-	-
16	F	35	Right maxillary swelling	Total resection	-
17	M	35	Left parietal swelling	-	68
18	F	52	Right frontal swelling	Total resection	-
19	F	15	Bilateral sphenofrontoethmoidal swelling	-	-
20	M	20	Nasal breathing problems	Biopsy	71
21	F	25	Right parietal swelling	-	-
22	M	18	Left exophthalmos and nasal breathing problems	-	-
23	F	27	Right parietal swelling	Total resection	25
24	F	20	Incidental (Right hearing loss)	-	-
25	M	20	Right frontoparietal swelling	Partial resection	-

***F:** Female, **M:** Male.*

Four different clinical forms of FD have been reported (12). The first form is MFD where only one bone is involved in body, and it accounts for about 70-80% of all FDs. Craniofacial bones are involved in 10-27% of MFD cases (9,10,12,23). MFD patients are generally in 20-30 years of age range. Lesions show no size increase after puberty. Solitary bone cyst, non-osteogenic fibroma, giant cell bone tumor, aneurysmal bone cyst, intraosseous meningioma, eosinophilic granuloma and adamantinoma should be considered in differential diagnosis of MFD (Figure 7A-F) (12,13). The second form is PFD involving multiple bones and it constitutes about 20-25% of all FDs. Craniofacial bones could be affected in 40-100% of all PFD cases (9,10,12). PFD generally manifests itself before the age of 10. These lesions increase in size during childhood ages and sometimes even after puberty. Hyperparathyroidism bone lesions, polyostotic Paget's disease, enchondromatosis and Cherubismshould be considered in differential PFD diagnosis (Figure 7A-F) (12). The third FD form is a component of McCune-Albright syndrome, which is observed in about 3% of FD patients. In this syndrome, PFD is accompanied by endocrine disorders such as hyperpigmented skin lesions in the form of light brown foci, puberty precox, hyperthyroidism, Cushing's disease, hyperprolactinemia and acromegaly. The fourth form of FD is Mazabraud syndrome in which intramuscular myxoma and PFD coexist (12,14). There are different classifications of craniofacial involvement of FD in literature. Some authors consider more than one bone involvement in craniofacial region as MFD (7,25), while others consider multiple bone involvement in craniofacial region as PFD (2,12). Cruz et al. mentioned that FD in which multiple bones of appendicular skeleton are involved is the real PFD form (7). Cruze et al. also mentioned the uncertainty about whether typical intramembranous ossification of skull bones is related to multiple contiguous bone involvement pattern in craniofacial FD (7). Hanifi et al. reported similar frequencies of single or multiple craniofacial bone involvement in their studies (12), whereas Atalar et al. reported single bone involvement in 53.1% of cases and multiple involvement in 46.9% (2). Chen et al. found that 30% of the FD cases had single bone and 70% had multiple bone involvement (5). In the present study, on the other hand, single bone was involved in majority of the FD patients (64.0%) and multiple bones were involved in

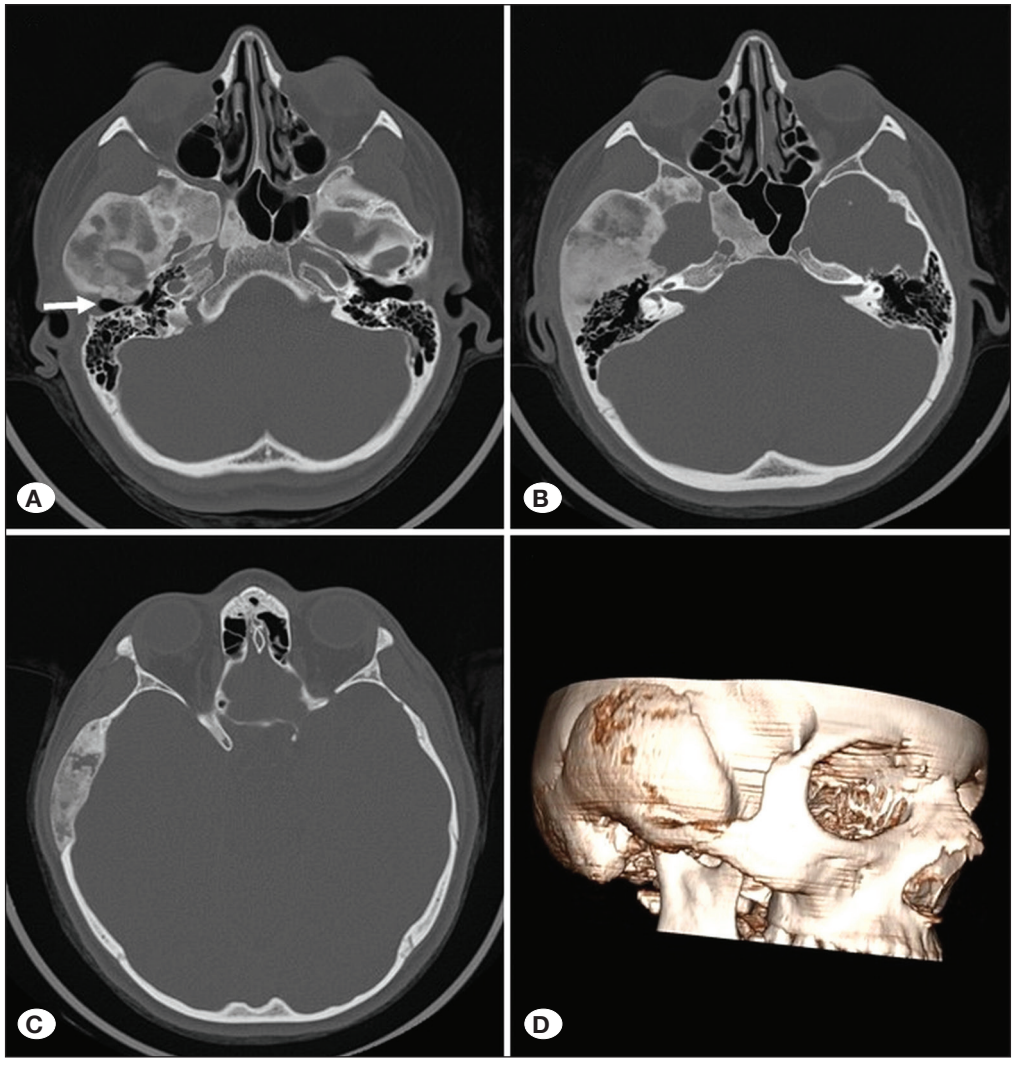

Figure 5: A 15-year-old male patient with complaints of narrowness in right outer ear canal and swelling in temporal region. **A-C)** FD lesion of mixed type (ground-glass + lytic + sclerotic) affecting right temporal and sphenoid bones and narrowing acoustic channel (white arrow) on axial plane CT slices. **D)** Swelling in lesion area and irregularities in cortical surface are shown on volume rendering CT image.

Table II: Radiological Findings of Patients with Fibrous Dysplasia

Patient No	Involved Bones	CT	MRI		
			T2-Weighted	T1-Weighted	+C
1	S	Ground-glass + sclerotic + lytic	-	-	-
2	M, Z, S	Ground-glass + sclerotic	-	-	-
3	M	Ground-glass + sclerotic	Heterogeneous hypointense	Heterogeneous hypointense	Severe heterogeneous
4	S, F, E, P	Ground-glass	-	-	-
5	P	Ground-glass + sclerotic	-	-	-
6	M	Ground-glass +sclerotic+lytic	-	-	-
7	M	Ground-glass + sclerotic	Heterogeneous mild hypointense	Heterogeneous mild hypointense	
8	T	Sclerotic	-	-	-
9	P	Ground-glass	Homogenous hypointense	Homogenous hypointense	-
10	M	Sclerotic	Homogenous hypointense	Homogenous hypointense	-
11	S, E	Ground-glass + sclerotic	Heterogeneous hypointense	Heterogeneous hypointense	Severe heterogeneous
12	Man	Ground-glass + sclerotic	-	-	-
13	F	Ground-glass + sclerotic	Heterogeneous hypointense	Heterogeneous hypointense	
14	S, T	Ground-glass + sclerotic + lytic	-	-	-
15	M	Ground-glass	-	-	-
16	M	Ground-glass + sclerotic + lytic	-	-	-
17	P	Ground-glass	Homogenous hypointense	Homogenous hypointense	
18	F	Ground-glass	Homogenous hypointense	Heterogeneous hypointense	Severe heterogeneous
19	S, F, E	Ground-glass + sclerotic + lytic	-	-	-
20	F, E	Ground-glass + sclerotic + lytic	-	-	-
21	P	Ground-glass	-	-	-
22	M, S, F	Ground-glass + sclerotic	-	-	-
23	P	Ground-glass	Mild hypointense	Mild hypointense	Increasing towards late phases
24	S, O	Ground-glass	-	-	-
25	S, F, P	Ground-glass + sclerotic	Hypointense	Heterogeneous hypointense	Severe heterogeneous

CT: *Computed tomography,* ***MRI:*** *Magnetic resonance imaging,* ***+C:*** *Contrast-enhanced,* ***E:*** *Ethmoidal,* ***F:*** *Frontal,* ***M:*** *Maxillary,* ***Man:*** *Mandibular,* ***P:*** *Parietal,* ***S:*** *Sphenoid,* ***T:*** *Temporal,* ***Z:*** *Zygomatic.*

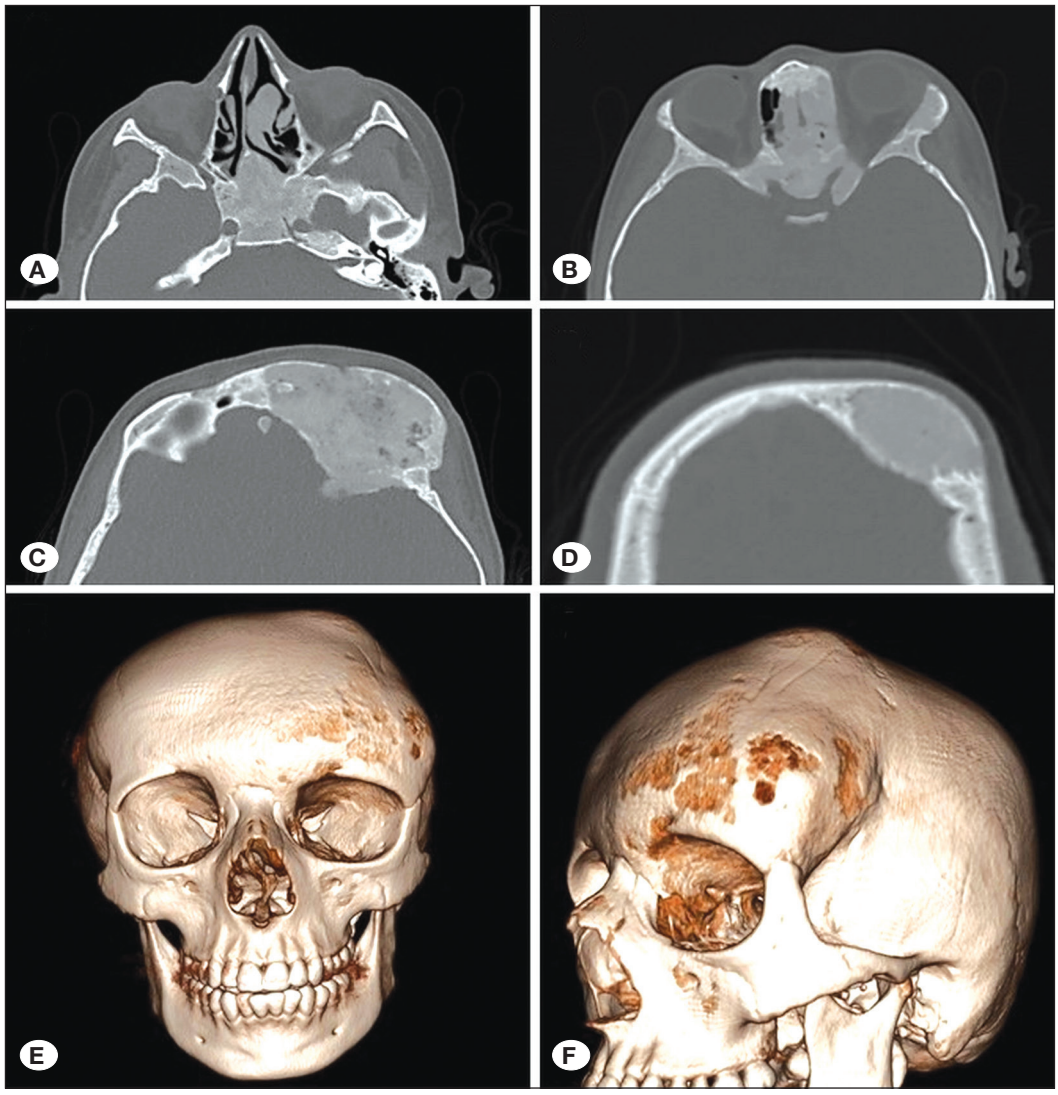

Figure 6: A 15-year-old female patient with swelling in cranium for a long time. **A-D)** Mixed type (ground-glass + sclerotic + lytic) FD containing mostly ground-glass densities and causing expansion in sphenoid bones and left frontal bone is shown on axial plane CT images. **E, F)** Dysmorphic, extended bone tissue stretching from left orbital roof to convexity plane is shown on volume rendering CT images.

others (36.0%). McCune-Albright and Mazabraud syndromes were not observed in any patient.

The symptoms of FD vary according to involved bones (18). FD lesions could lead to neuropathy, obstruction or loss of function through neighborhood to or compression of vital structures (22). However, a slow-growing painless swelling is most common symptom (18). Depending upon the involved area, symptoms such as (hearing or visual loss, headache, proptosis, exophthalmos, dystopia, hypertelorism, facial pain, trigeminal neuralgia, tinnitus, nasal obstruction, sinusitis symptoms, anosmia, malocclusion, teeth loss, cranial asymmetry and facial deformity) could be seen within years (7,12,19,22,24). Seventeen patients (68.0%) in the present study had hard swelling complaint depending upon the involved area. Hard swelling complaint was accompanied by narrowing in external acoustic channel in one patient and visual loss in another. In terms of complaints of patients, one patient (4.0%) had visual loss and trigeminal neuralgia in affected side, one patient (4.0%) had chronic otitis media, one patient (4.0%) had headache and nasal breathing difficulty, one patient (4.0%) had exophthalmos and nasal breathing difficulty, one patient (4.0%) had nasal breathing difficulty only, while lesions were incidentally found in three patients (12.0%) in examinations performed because of other etiologies. Six of the patients in our series were operated. Five of them were operated for cosmetic purposes due to swelling in involved area, while the sixth was operated because of swelling and loss of vision.

CT is best imaging modality to evaluate radiographic characteristics and boundaries of FD lesions (4,22). Opacity in ground-glass density, ballooning and expansion and thinning in cortex are characteristics of FD in CT (19). Based on the amounts of their components, FD lesions could have different appearance patterns in CT such as ground-glass, sclerotic or homogenous dense, mixed or pagetoid and cystic. Appearance patterns of FD lesions in CT could also vary depending upon the age of patient (4,22). Lesions on CT are mostly homogeneous and radiodense in the first decade or in pre-pubertal period of life. Towards the second decade of life, lesions turn to a mixed radiodense/radiolucent appearance

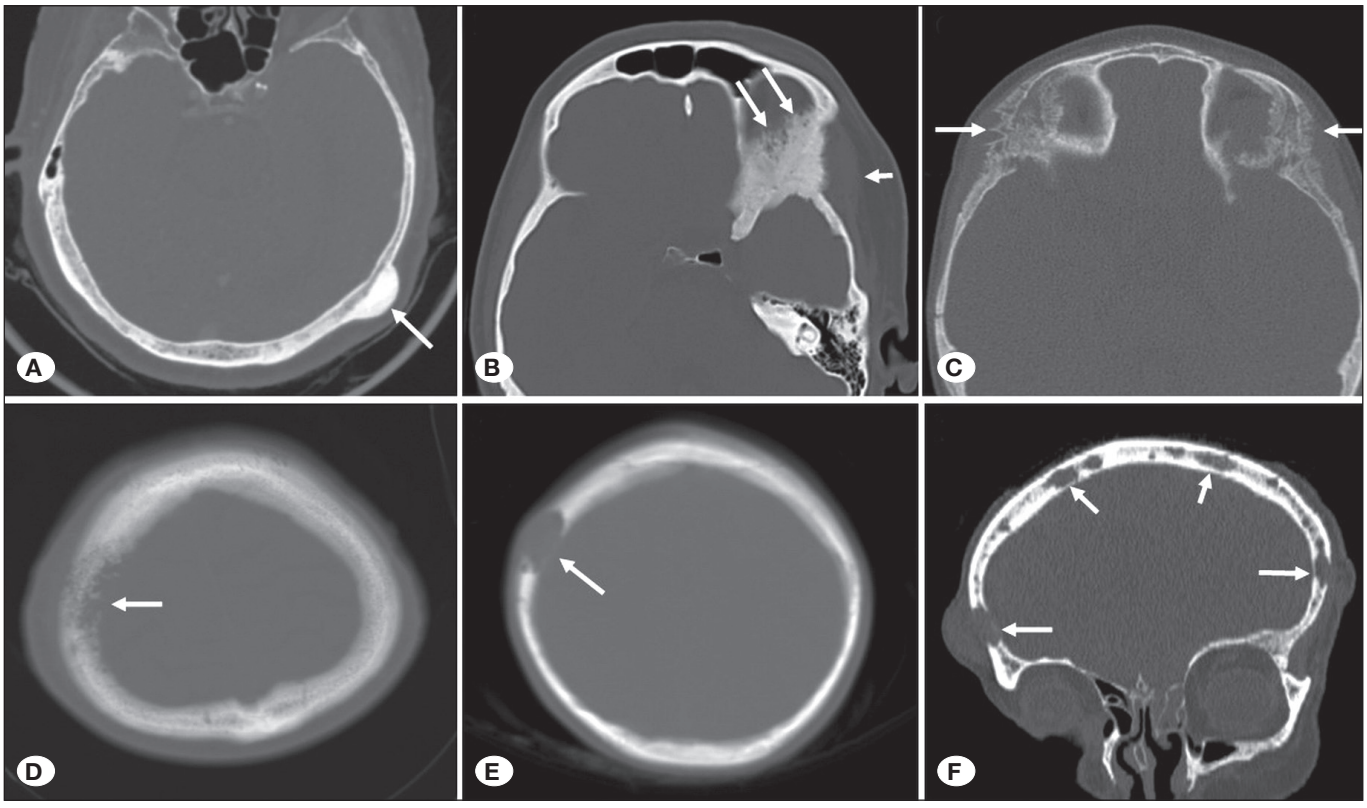

Figure 7: CT images of some lesions that could be used in differential diagnosis of fibrous dysplasia. **A)** Homogeneous hyperdense osteoma (arrow) is observed on left occipital bone of a 69 years old female patient. The fact that lesion originated from outer tabula and without involved diploe allows its differentiation from fibrous dysplasia. **B)** Forty-nine years old female patient with intraosseous meningioma located in left orbital wall. Radial bone spiculation of contours (long arrows) and presence of accompanying soft tissue component (short arrow) differentiates the lesion from dysplasia. **C)** Metastases (arrows) leading to radial bone spiculation in bilateral orbita in a one year old female patient with neuroblastoma on bilateral orbital bones. **D)** Sixty-four years old male patient with bladder cancer metastasis on right parietal bone. Irregularly contoured lytic areas in the bone (arrow) and accompanying soft tissue component differentiates it from fibrous dysplasia. **E)** Six years old female patient with eosinophilic granuloma shows osteolytic lesion on right parietal bone (arrow). **F)** Multiple lytic lesions (arrows) accompanied by scattered soft tissues in cranial bones of 65 years old female patient with multiple myeloma are shown.

and are stabilized in adulthood, some of them turning to a ground-glass appearance (4,22). Turning of FD lesions to mixed pattern alone is not an indication for a more detailed examination or biopsy, and CT follow-ups are recommended for these patients (4). Most of the patients in the present study (60.0%) were mixed type (ground-glass + sclerotic and ground-glass + sclerotic + lytic components), while only two (8.0%) were pure sclerotic type FD. Because of lack of follow-up radiological images for the patients under 20 years of age in the present study, these patients were not evaluated for pattern changes. No pattern and clear size changes were observed in lesions of seven patients over 20 years of age who were monitored with follow-ups.

MRI findings of FD could be confusing (1,16). Lesions are characterized by decreased signal along with sharply defined borders on T1- and T2-weighted images (4). On MRI, FD does not exhibit its distinctive features in radiography or CT, and often resembles the characters of tumors (4). This is especially true when lesion has intermediate signal intensity on T1-weighted images, high intensity on T2-weighted images and clear contrast-enhancement with the injection of contrast matter. Likelihood of correct diagnosis of FD with MRI is high only when signal intensities of both T1- and T2-weighted images are low despite injection of contrast matter (4,6). In the present study, FD lesions had homogeneous or heterogeneous hypointense signal features on T1- and T2-weighted MRI images. No diffusion restriction was observed on diffusion-weighted MRI series. Dense heterogeneous contrasting was observed in lesions of four patients who had contrast-enhanced MRI, while in another patient contrast-enhancement started as focal contrast-enhancement areas in early phases and turned to intense contrast-enhancement in late phase (Figure 4A-F).

The present study has several limitations. First, lesions of only nine patients had histological diagnoses, and lesions of other patients were diagnosed only by radiological methods. Second, since follow-up examinations were carried out only on seven patients, possible pattern changes were not evaluated. Third, since only ten patients had MRI examination, imaging findings from CT and MR examinations of all lesions could

not be compared. Finally, although the present study included one of the largest series in literature, there was relatively fewer cases and some FD types (e.g. pure cystic lesions) did not exist in our patient population.

COCNLUSION

In conclusion, craniofacial FD is more common in young adults and females. It often causes swelling in involved bones and/or findings of pressing on adjacent structures. It usually involves single bones, frequently maxillary or sphenoid bones. Depending upon its components, craniofacial FD could manifest different imaging features on radiological examinations.

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