



## A retrospective study on craniofacial fibrous dysplasia: Preoperative serum alkaline phosphatase as a prognostic marker?



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### ABSTRACT

**Background:** Craniofacial fibrous dysplasia (CFD) often requires surgery to correct facial deformity and prevent functional impairment. However, recurrence is common, and there is no reliable prognostic biomarker. The aim of this paper is to evaluate the possibility of using preoperative alkaline phosphatase (ALP) as a prognostic marker for CFD.

**Material and methods:** Forty-nine patients with CFD who underwent surgery from 2000 to 2011 were selected. The relationship between preoperative ALP and age, gender, lesion type and prognosis was investigated.

**Results:** The recurrence rate was 31.8% in patients who received conservative bone contouring. Patients with recurrence did not show significantly higher levels and abnormal rates of ALP than patients without recurrence. Young patients and those with polyostotic CFD showed higher ALP levels than adults and those with monostotic CPD ( $P < 0.05$ ). Although CFD patients showed higher levels and abnormal rates of ALP than the control group, significant levels were not reached ( $P > 0.05$ ). No correlation between age, gender, type, ALP and recurrence could be established using the logistic regression model.

**Conclusion:** Preoperative ALP may not be a reliable prognostic marker of CFD based on the findings in this study. Close follow-up is recommended after conservative bone contouring.

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### 1. Introduction

Fibrous dysplasia (FD) was first reported by Lichtenstein in 1938 (Lichtenstein, 1938). It is rare benign bone disorder characterized by the replacement of normal bone and marrow with fibrous tissue intermixed with irregular woven bone. FD accounts for 3% of all bony tumours and over 7% of all non-malignant tumours of bone (Edgerton et al., 1985; Yabut et al., 1988).

The aetiology of FD is related to the mutation of gene (GNAS 1) encoding the alpha-subunit of a signal-transducing G-protein. The mutation of GNAS 1 keeps adenylyl cyclase persistently active, thus producing increased cAMP activity leading to hyperfunction of skeletal progenitor cells and abnormal osteoblasts (Cohen and Howell, 1999). Increased interleukin-6 secretion, one of the

downstream effectors of cAMP, may also take part in the process of FD by increasing the number of osteoclasts (Yamamoto et al., 1996).

FD usually begins in childhood and typically manifests as a progressive painless swelling. It can involve a single bone (monostotic FD; MFD), or multiple bones (polyostotic FD; PFD). Rarely PFD may present with café-au-lait skin macules and endocrinopathies. This is known as the McCune–Albright syndrome (Albright et al., 1937). The most commonly affected sites are the craniofacial skeleton, the proximal femur, rib, and tibia. Nearly 50% of patients with PFD and 10–27% of patients with MFD have craniofacial skeleton involvement. Affected bones include the maxilla, mandible, sphenoidal, zygomatic, frontal, ethmoidal, parietal, temporal and occipital bones. The commonest symptom of craniofacial FD (CFD) is severe facial deformity (Assaf et al., 2012; Boni et al., 2011; Keskin et al., 2009) but CFD can also cause other symptoms, such as dysaesthesia, nasal obstruction, proptosis and loss of vision or hearing depending on the sites of the skeleton affected (Ricalde and Horswell, 2001).

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The main treatment for CFD is surgery, which can be divided into conservative and radical resection. Conservative bone contouring is most commonly used to improve facial aesthetics and prevent functional impairment. Conservative bone contouring does however have a high recurrence rate ranging from 25% to 82% and repeat surgery is often inevitable (Edgerton et al., 1985; Kusano et al., 2009).

With the potential possibility of recurrence deciding the timing and extent of surgery is challenging. A sensitive and reliable prognostic marker may be of great help when planning surgery (Lee et al., 2012). Alkaline phosphatase (ALP) is an important hydrolase enzyme related to osteoblastic activity. It has been used as a prognostic marker for bone-related tumours (Bacci et al., 1993; Liu et al., 1996). Patients with FD often show higher ALP levels. Following medical treatment of FD with diphosphate ALP levels decrease along with reduced bone destruction and ALP levels have been used to monitor treatment response (Lala et al., 2000; Chapurlat et al., 2004; Chen et al., 2006). ALP levels are also often higher in cases of malignant transformation of CFD (Sadeghi and Hosseini, 2011). All these characteristics make ALP a promising prognostic marker of CFD.

Forty-nine patients with CFD who received surgery in our hospital from 2000 to 2011 were identified. Another 49 patients without CFD matched by age and gender were chosen as the control group. The preoperative serum ALP levels in patients of different ages, gender and lesion types were compared and the relationship between ALP level and recurrence was investigated. The clinical features of CFD are also discussed.

## 2. Material and methods

### 2.1. Patients

This retrospective study was approved by the ethics committee of the General Hospital of the People's Liberation Army. A total 49 patients with CFD who received treatment at the Department of Oral and Maxillofacial Surgery, Chinese PLA General Hospital (Beijing, China), between 2000 and 2011 were included. Twenty-two of the patients were followed up for at least a year. A diagnosis of CFD was confirmed by postoperative pathological examination in all patients. Patients with McCune–Albright syndrome were not included in this study. Forty-nine patients hospitalized for dental extractions, matched by age and gender, were randomly chosen as the control group.

The reference values of ALP for young patients ( $\leq 18$  years) are listed in Table 1. For adults ( $> 18$  years) levels of 20–140 U/L were adopted as the reference range.

### 2.2. Statistics

Patients were divided into different subgroups based on gender, age, type of CFD, and prognosis. The mean preoperative ALP level between different subgroups was compared using the *t*-test. The rate of abnormal ALP and the distribution of recurrence among

**Table 1**  
The reference values of ALP for children.

Age	Value (U/L)	
1–9 years	145–420	
10–11 years	140–560	
	Male	Female
12–13 years	200–495	105–420
14–15 years	130–525	70–230
16–19 years	65–260	50–130

According to Nelson textbook of pediatrics, 19th edition (Stanley, 2010).

**Table 2**  
General characteristics of patients with CFD between 2000 and 2012 ( $n = 49$ ).

	N	Percent (%)
Age of onset		
1–9 years	10	20.4
10–18 years	23	46.9
>18 years	16	32.7
Gender		
Male	28	57.1
Female	21	42.9
Type		
MFD		
Mandible	20	40.8
Maxillae	9	18.4
PFD	20	40.8
Symptoms		
Deformity	49	100
Pain	15	30.6
Numb	3	6.1

subgroups was compared using the Chi-square ( $\chi^2$ ) test. The logistic regression model was used to assess factors that may be related to recurrence such as age, gender, type, and ALP level. All the analyses were performed using SPSS 13.0 (SPSS, Inc, Chicago, IL), and  $P < 0.05$  was considered statistically significant.

## 3. Results

### 3.1. General characteristics

The general characteristics of the study population are summarized in Table 2. Twenty-eight patients were male and 21 were female (male to female ratio 1.3:1). The age of onset ranged from 2 to 62 years old (median 14 years).

Twenty-nine patients presented with MFD (59.2%) and 20 with PFD (40.8%). In MFD patients the mandible was the most commonly affected bone (74.1%). The maxilla was affected in 9 patients (25.9%). In patients presenting with PFD affected bones included the zygomatic, sphenoidal, frontal and temporal bones.

Progressive maxillofacial deformities were observed in all patients and this was the main reason for seeking treatment. The second most common symptom was facial pain and 15 patients complained of this. Other symptoms included numbness (6.1%) and one patient suffered from hearing loss and visual disturbance.

### 3.2. Surgery and recurrences

The age at the time of the first surgery ranged from 3 to 63 years, with a median age of 22 years. Most patients received conservative bone contouring with different approaches (44/49). In 5 patients with MFD complete resection of the mandibular lesion with immediate reconstruction using an iliac bone graft was carried out. Lefort I osteotomy was performed in a patient with PFD involving the maxilla and the zygomatic bone.

Recurrences occurred in 7 (31.8%) out of 22 patients who had regular follow-up. All 22 patients had conservative bone contouring. The distribution of recurrences is listed in Table 3. Male

**Table 3**  
The recurrence rate in different subgroups ( $n = 22$ ).

	Age		Gender		Type	
	Young	Adult	Male	Female	MFD	PFD
Recurrence rate	3/8 (37.5%)	4/14 (28.6%)	5/14 (35.7%)	2/8 (25%)	3/12 (25%)	4/10 (40%)
<i>P</i> value ( $\chi^2$ -test)	0.665		0.604		0.452	

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