

PERIPHERAL ODONTOGENIC FIBROMA IN A TAIWAN CHINESE POPULATION: A RETROSPECTIVE ANALYSIS

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A retrospective analysis of 25 cases of peripheral odontogenic fibroma (World Health Organization-type) (PODF [WHO-type]) in a Taiwan Chinese population was performed at a single institution. The clinical findings, which included a wide age distribution, a female preponderance and no racial predilection, were consistent with those of previous case series reports; however, a slight preference for location in the maxilla was found in the current case series. It is noteworthy that one particularly sizable lesion was identified in a 30-year-old Chinese female who presented with a swollen mass in the right mandible that had been present for about 2 years. Intraoral examination revealed an exophytic firm mass that measured 4.5 × 4 cm, located over the right mandibular edentulous ridge from the second premolar to the second molar area. Both panoramic radiography and computerized tomography revealed multiple radiopacities within the tumor. In conclusion, the analysis of clinical data in the current case series of PODFs (WHO-type) occurring in Taiwan Chinese mostly corroborates other reports; however, an unusually large mandible lesion was noted, indicative of the apparent size that such tumors can reach unless they are surgically removed at an early stage.

Key Words: Chinese, large, mandible, peripheral odontogenic fibroma
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Peripheral odontogenic fibroma (PODF) is an uncommon dental lesion regarded by the World Health Organization (WHO) as a benign odontogenic neoplasm derived from fibroblasts [1]. Gardner [2] was the first to define PODF, which was initially thought to be a rare extraosseous counterpart to central odontogenic fibroma and distinct from peripheral ossifying

fibroma [3]. PODFs are more prevalent in females in the second decade of life [2,4], and there is a trend towards occurrence in the posterior mandible [5]. PODF usually presents clinically as either a pedunculated or a sessile mass that is similar in color to the surrounding connective tissue and evolves as a slow-growing, non-ulcerated, asymptomatic exophytic lesion attached to the gingiva [6]. Histologically, PODF is characterized by the presence of islands of odontogenic epithelia and either no calcification or dentinoid/cementum-like material associated with it [5]; they are usually small, with an average diameter of approximately 1–2 cm [7]. To our knowledge, there has never been a case series study of PODFs (WHO-type) in a Taiwan Chinese population. The aim of the current study was therefore to present a retrospective



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Table 1. Clinical data relating to the 25 cases in the present series

Case	Age (yr)	Sex	Gingival location	Maximum diameter (cm)	Clinical diagnosis
1	65	M	Maxilla, 1 st to 2 nd premolar, right, buccal	1.5	PG
2	32	M	Maxilla, 1 st premolar, right, buccal	1.0	PODF
3	35	M	Mandible, central to lateral incisors, right, labial	1.0	F
4	60	M	Maxilla, 1 st to 2 nd premolar, right, buccal	1.5	PODF
5	18	F	Mandible, lateral incisor to canine, left, labial	0.7	F
6	44	F	Mandible, lateral incisor to canine, left, labial	0.9	PG
7	35	M	Mandible, central incisors, bilateral, palatal	1.0	F
8	47	F	Mandible, 2 nd molar, left, buccal	2.0	F
9	59	M	Mandible, central incisors, bilateral, labial	0.5	F
10	14	F	Maxilla, lateral incisor to canine, left, labial	1.9	PODF
11	28	F	Maxilla, 1 st to 2 nd premolar, left, buccal	1.5	PG
12	40	M	Maxilla, central to lateral incisors, left, labial	0.6	PG
13	27	M	Maxilla, central to lateral incisors, right, palatal	1.2	PG
14	37	F	Mandible, central incisor to canine, left, palatal	1.3	PG
15	23	F	Maxilla, central to lateral incisors, left, labial	1.0	PODF
16	27	M	Maxilla, 1 st molar, right, buccal	0.6	PG
17	37	F	Maxilla, canine to 1 st premolar, right, buccal	1.3	PG
18	25	F	Maxilla, 1 st to 2 nd premolar, right, palatal	1.7	PG
19	54	F	Mandible, lateral incisor to canine, left, labial	1.0	F
20	24	F	Mandible, 2 nd premolar, right, lingual	0.5	F
21	26	F	Mandible, 2 nd premolar to 1 st molar, right, lingual	1.0	PG
22	NA	F	Maxilla, lateral incisor to canine, right, labial	1.0	F
23	55	F	Maxilla, canine, right, labial	1.4	PODF
24	30	F	Mandible, edentulous 1 st to 2 nd molar, right	4.5	PODF
25	45	F	Maxilla, 2 nd premolar to 1 st molar, left, buccal	0.5	PG

PG = pyogenic granuloma; PODF = peripheral odontogenic fibroma (WHO-type); F = fibroma; NA = not available.

analysis of a cohort of Taiwan Chinese patients diagnosed with PODF (WHO-type) in a single institution, and compare the results with those of previous studies. In addition, we present an unusual case history of a 30-year-old woman with a particularly sizable lesion that occurred in the mandible.

METHODS

The patient database of the Oral and Maxillofacial Pathology Department of our institution was searched for all patients diagnosed with PODF between January 2000 and January 2007, and the records were retrieved and the cases reviewed microscopically in accordance with the WHO classification [1]. Twenty-five cases were identified as being suitable for retrospective analysis. Clinical information relating to the patients, including age, gender, lesion location, size and clinical diagnoses, was obtained from the biopsy submission forms and clinical charts. Reliable clinical data relating to Malay (20 cases) and Malay Chinese (12 cases)

populations were taken from the study by Siar and Ng [8] for comparison, and data relating to Caucasians (18 cases) were identified from a report by Buchner and colleagues [9].

RESULTS

The clinical features of the 25 cases of PODF (WHO-type) in Taiwan Chinese patients are summarized in Table 1. The youngest patient was a 14-year-old girl and the oldest was a 65-year-old man; the mean age at diagnosis was 37 years. PODF occurred in nine (36%) males and 16 (64%) females. Peak incidence was in the second to third decades of life (52%). The most frequent location was the maxilla (14 cases, 56%), while the PODF occurred in the mandible in 11 cases (44%). The mandibular incisor/canine and premolar areas were the most predominant sites, affecting both the buccal/labial and lingual/palatal aspects of the gingiva. All lesions were non-encapsulated, and calcified odontogenic epithelium (dentinoid

Table 2. Comparison of clinical demographic data in the present case series with those of studies on other ethnic origins [8,9]

Patients' characteristics	Racial distribution			
	Taiwan Chinese	Malay Chinese*	Malay*	White [†]
Male/Female	9/16	7/5	9/11	10/8
Age range (yr)	14–65	11–64	4–64	12–84
Mean age (yr)	37	33	33	34.2
Maxilla/mandible	14/11	5/7	8/10 [‡]	5/13
Total number of cases	25	12	20	18

*Data derived from reference 8; [†]data derived from reference 9, in which 18 cases were white, three cases were black, one was Hispanic and one was Asian; [‡]two cases of unknown site.

and/or cementum-like material) was present in all cases. Fifteen (60%) cases revealed various degrees of radio-opaque shadows on radiographic examination. Twenty-one cases were initially diagnosed as having different conditions on clinical grounds, these being pyogenic granuloma (13 cases) and fibroma (8 cases). All cases were treated with simple surgical excision and no recurrence was noted. Furthermore, when comparing the clinical demographic data with the results of studies of other ethnic populations (including Malay Chinese, Malay and Caucasian), the only exception identified was a greater frequency of lesions located in the maxilla. This information is summarized in Table 2 [8,9].

All lesions measured between 0.5 cm and 2 cm with the exception of one (case no. 24, Table 1), which was found to be 4.5 × 4 cm. This unusual case occurred in a 30-year-old female who was referred to our institution for evaluation of a swollen mass in the right mandible. The patient explained that the mass had been present for about 2 years and had started to enlarge over the previous 4 months. The first and second right mandibular molars had exfoliated due to increased mobility as a result of the growth of the mass. Extraorally, mild facial asymmetry was noted due to the bulging of the right cheek. Intraoral examination revealed an exophytic mass of gingiva-like tissue that occupied the right posterior mandibular edentulous ridge, extending from the second premolar to the second molar area (Figure 1). The occlusal surface of the mass was extensively ulcerated due to repeated trauma from the upper teeth (Figure 1). The right mandibular second premolar was shifted to the lingual side by the mass. Mild tenderness was also noted and the mass was firm in consistency. Panoramic radiography revealed a well-defined unilocular

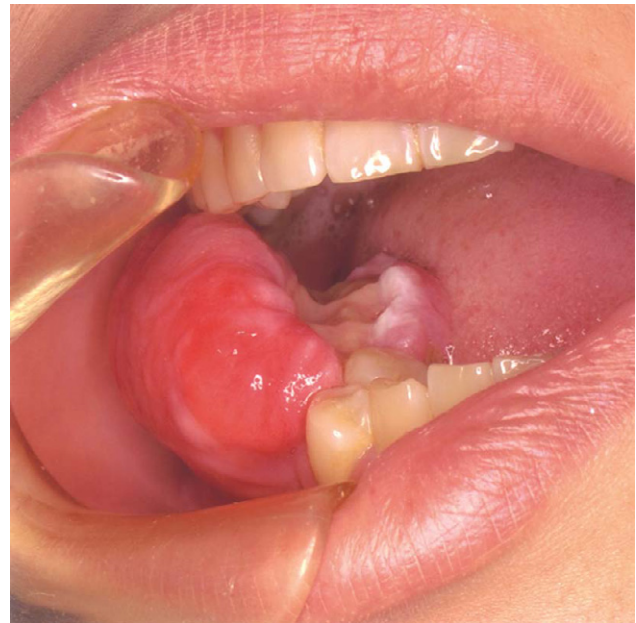


Figure 1. An exophytic mass occupying the edentulous ridge between teeth 45 and 47.

boat-shaped radiolucency with multiple radio-opacities over the right mandibular body. The lesion extended from the distal aspect of the mandibular second premolar to the mesial aspect of the residual root of the mandibular third molar, and from the inferior cortical crest down to the apex of the mandibular second premolar (Figure 2). Computed tomography showed a round-shaped soft tissue mass with some calcifications in the right mandibular body, extending from the distal side of the mandibular second premolar to the mandible ridge, associated with bony destruction to the upper third of the inferior cortical bone (Figures 3A & B). The patient admitted to occasional cigarette smoking and alcohol consumption, but denied betel nut chewing. The entire mass was subsequently surgically

removed under general anesthesia, together with the lingually displaced lower right second premolar. The extraction of all residual roots of the right lower third molar, left lower first to third molars, and left upper third molar, as well as the deep-decayed right upper third and left upper first molars, was planned during surgery; however, the distal root of the left lower first molar was accidentally retained and was removed on a subsequent follow-up visit. Furthermore, during surgical excision, the tumor mass was found to be pedunculated (Figure 4), and was attached to the alveolar mucosa between the right mandibular second premolar and the third molar, demonstrating that the lesion was not an extrasosseous counterpart of a central odontogenic fibroma. The histologic findings are shown in Figure 5. The patient has remained free of recurrence during a follow-up period of more than 12 months.



Figure 2. Panoramic radiography reveals a well-defined, unilocular boat-shaped radiolucency with multiple radio-opacities over the right mandibular body.

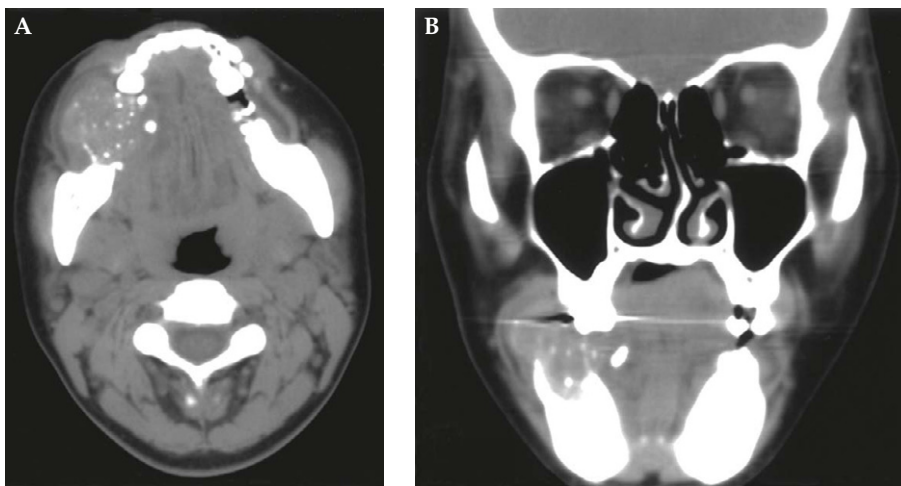


Figure 3. Computed tomography demonstrates a round, soft tissue mass with some calcifications in the right mandibular body and bony destruction downward to the upper third of the inferior cortical bone.

DISCUSSION

There are several retrospective studies on PODFs (WHO-type) already in the literature [4,5,7–10]. Garcia and colleagues [7] observed a much higher prevalence of these lesions (17 cases) in females (88.2%), that they occurred most frequently in the second to the fourth decades of life (76.4%), and that they were predominantly located in the mandible (58.7%). Daley and Wysocki [10] found that the 36 cases in their study had a wide age range (varying from 12 to 75 years), and reported a greater incidence in females (male:female ratio, 1:2) and a slight predominance of location in the mandible (55.6%). Siar and Ng [8] reported on 46 cases and indicated a slight female preponderance (male:female ratio, 1:1.3), a peak incidence in the



Figure 4. Macroscopic view of the surgical specimen of the excised pedunculated (indicated by a star) tumor.

second decade of life, with a wide age range from 5 months to 64 years, and a slight bias towards location in the mandible (52.2%). Buchner and colleagues [9] again demonstrated a slight female preponderance (male:female ratio, 1:1.1), with most cases occurring in the second to the fourth decades of life (82.6%), a wide age range from 12 to 84 years, and a preference for location in the mandible (69.6%, 23 cases). Kenney and colleagues (13 cases) [5] noted a more frequent occurrence in the mandible (69.3%) and a peak incidence in the third to fourth decades of life (38.3%), but a slight male predominance (male:female ratio, 1.6:1). Finally, Slabbert and Altini [4] also reported a higher prevalence in men (male:female ratio, 1.3:1) and a wide age distribution (11–76 years), with most cases occurring in the third decade of life, and a slight predominance of location in the mandible (52%, 30 cases). Taken together, the clinical findings from our case series (Table 1) compare favorably with those of

previous case series [4,5,7–10], including a wide age distribution, a female preponderance and no racial predilection (Table 2). However, in contrast to previous studies, our case series analysis revealed a slightly higher frequency of lesions located in the maxilla [4,5,7–10].

Odontogenic epithelium with calcification (dentinoid and/or cementum-like material) was present in all cases of the present case series; however, not all cases (60%) showed radio-opaque shadows on radiographic examination. Therefore, when imaging a lesion, a small modification of exposure factors, such as a decrease in voltage and an increase in current, may be required in order to show tiny radio-opaque foci. In contrast to our finding of 100% presence of histologic calcification, mineralization was found to be present in 38.8% (14/36 cases) [10], 41.2% (7/17 cases) [7], 50% (23/46 cases) [8], 52% (12/23 cases) [9], and 73.3% (22/30 cases) [4] of cases in five previous studies.

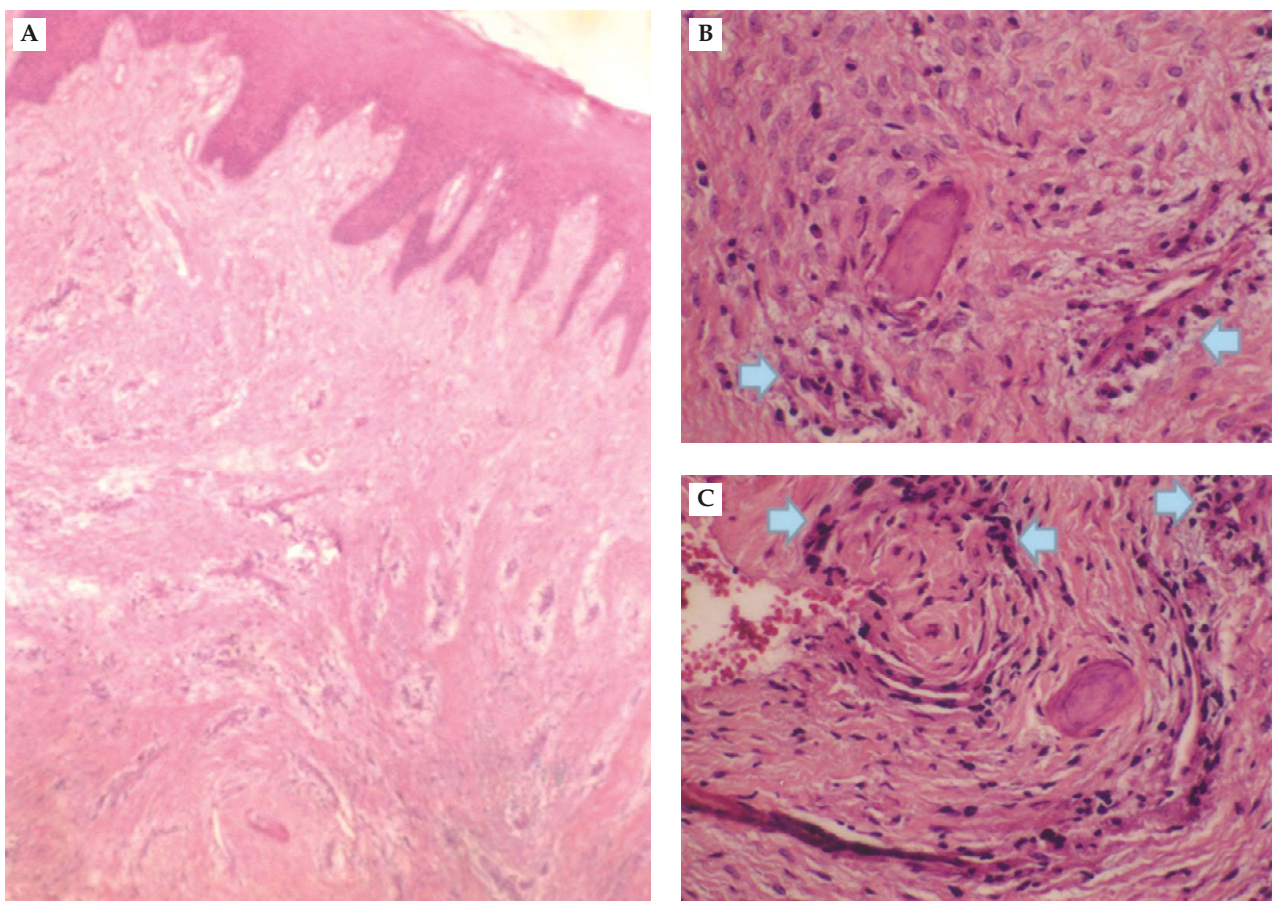


Figure 5. (A) The lesion is characterized by dense, cellular fibrous connective tissue and was found to be covered by stratified squamous epithelium on low-power examination (hematoxylin & eosin, 40 \times). (B, C) Small blood vessels, rests of odontogenic epithelium (arrows) and cementum-like substances were found on higher magnification (hematoxylin & eosin, 200 \times).

It is noteworthy that in the present case series, one particular lesion was so large that it caused tooth exfoliation and tilting, alveolar bone resorption, adjacent bony structure destruction and even facial asymmetry. In the most recent analysis by Garcia and colleagues [7], most lesions were found to be less than 2.0 cm in diameter. In another review by Buchner and colleagues [9], the diameter ranged from 0.5 cm to 1.4 cm. Finally, in the study by Slabbert and Altini [4], the lesions measured between 1.0 cm and 3.0 cm in diameter. To our knowledge, there has never been a report of a lesion larger than 3 cm. Thus, the present case series supports the true growth potential of this pathological entity.

In the current study (Table 1) as well as in previous case series [7–9], many lesions were clinically misdiagnosed as other types of lesions, such as peripheral ossifying fibroma (POF), pyogenic granuloma or peripheral giant cell granuloma. These may have similar clinical appearances and courses; hence, clinical findings alone are not sufficient for a diagnosis of PODF (WHO-type), and biopsy and histopathologic examination are necessary for a definitive diagnosis.

Histologically, the lesion that is most similar to PODF (WHO-type) is POF. Pindborg and colleagues [11] defined PODF as “a fibroblastic neoplasm containing varying amounts of odontogenic epithelium where the fibrous connective tissue component is mature and often contains osteoid, osteodentin or cementum-like material”. The main histologic difference between these two pathological entities is the presence of odontogenic epithelium and dysplastic dentin and/or cementum-like material in the PODF. It should be noted that the term “peripheral odontogenic fibroma” has been used to describe “peripheral ossifying fibroma” by some pathologists, but this should be avoided as PODF has been designated by the WHO as the rare and extraosseous counterpart of the central odontogenic fibroma [1]. Although the source of the epithelium is unknown, the predilection for the posterior mandible seems reasonable given that a high percentage (46.2%) of the normal opercula contains remnants of the odontogenic epithelium [12].

Extensive ulceration is seldom present in PODFs (WHO-type); however, extensive superficial ulceration due to occlusal trauma was noted in our patient. The recurrence rate for PODF is low: in Kenney et al’s report [5], only one case (7.7%) recurred. Therefore, simple surgical excision is the preferred treatment.

However, Daley and Wysocki [10] noted that seven out of 30 cases recurred, while Garcia and colleagues [7] found that three out of 17 cases recurred. Due to the clear growth potential of these lesions as seen in the present case, close follow-up should be adopted.

In conclusion, clinical data from a cohort of PODF (WHO-type) patients in a Taiwan Chinese population mostly corroborate the results of other case series. It should be noted that a particularly large lesion was identified in the current series, indicating the apparent growth potential of these lesions.

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發生於台灣人口的周邊性齒源性纖維瘤 之回顧分析

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本篇研究為針對單一學院之 25 例台灣人口所發生之周邊性齒源性纖維瘤 (世界衛生組織類型) 所做的回溯性分析。其臨床發現和過去所發表之案例相符合, 包含廣泛的年齡層分佈, 好發於女性, 且沒有種族偏好。然而, 在本篇研究呈現之病例可發現發生位置稍偏好於上顎。值得注意的是, 其中有一體積特別巨大的案例, 發生於一 30 歲中國女性, 據其自述。此發生於右側下顎之腫塊已有兩年之久。口內檢查顯示出在右下顎第二小白齒至第二大白齒之無牙脊區域有一直徑約 4.5×4 公分大小之外生性堅硬腫塊。於齒顎全景 X 光線攝影及電腦斷層掃描皆顯示出腫瘤內有多發性放射線不透區。結論, 根據本篇關於台灣人口所發生之周邊性齒源性纖維瘤 (世界衛生組織類型) 病例之臨床資料分析結果, 多數可和其文獻報告相互印證。然而, 一罕見之巨大下顎病灶已被發表, 表示此腫瘤有生長變大之可能, 除非於發病早期即以手術摘除。

關鍵詞: 台灣人, 巨大, 下顎, 周邊性齒源性纖維瘤
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