Case report

Case report of intra-osseous fibroma: a study on odontogenic and desmoplastic fibromas with a review of the literature

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Abstract: Intra-osseous fibromas of the jaw are classified by origin. Intra-osseous odontogenic fibromas have odontogenic epithelia, while desmoplastic fibromas do not. However, it is often difficult to determine the odontogenic origin for central fibromas. Three subjects with a diagnosis of intra-osseous fibroma were examined. Case 1 was a 35-year-old man found to have a panoramic radiograph from the right premolar to the mandibular ramus in the mandible that exhibited multilocular radiolucency. Within the radiolucency, small-radioopaque bodies were observed. Case 2 was a 13-year-old female, in whom a panoramic radiograph from the left premolar to the molar in the mandible showed multilocular radiolucency. Case 3 was a 51-yearold female who exhibited a heart-shaped radiolucency in the panoramic radiograph of the left first molar area in the mandible. We also reviewed the literature for previously reported cases of intra-osseous odontogenic and desmoplastic fibroma. In 64 cases of intra-osseous odontogenic fibroma and 68 cases of desmoplastic fibroma we extracted data on age, sex, location, and radiographic findings. Based on the analysis of the reported literature cases, re-evaluation of the patients in our study revealed that case 1 could be classified as desmoplastic fibroma, while cases 2 and 3 were intra-osseous odontogenic fibromas. (J. Oral Sci. 47, 149-157, 2005)

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Introduction

Fibromas (WHO type) often occur in the oral mucosa, but certain rare cases have been reported to involve the jawbone (intra-osseous fibroma). Depending on the presence or absence of odontogenic epithelia, intra-osseous fibroma cases can be classified into two respective categories: odontogenic fibroma or non-odontogenic fibroma (desmoplastic fibroma) (1-4). The clinical findings for intra-osseous odontogenic fibroma are similar to those seen with desmoplastic fibroma in the jaws. However, desmoplastic fibroma is generally believed to be more aggressive than odontogenic fibroma(1-3).

In this paper, we describe three cases of intra-osseous fibroma. In addition, we also reviewed the literature for cases of intra-osseous odontogenic and desmoplastic fibroma. Clinical findings from these cases were extracted in order to determine the difference, if any, between clinical findings for intra-osseous odontogenic and desmoplastic fibroma.

Materials and Methods

Three patients were diagnosed with intra-osseous fibroma at a hospital affiliated with the Nihon University School of Dentistry at Matsudo. The age and sex, as well as the location and radiographic findings for the fibromas were obtained for each of the patients. CT image findings were also added to the patient profiles.

Our literature review found 64 cases of intra-osseous odontogenic fibroma (29 reports) (5-33) and 69 cases of

desmoplastic fibroma (43 reports) (34-76). After extracting the data for age, sex, location, and radiographic findings, we attempted to determine if differences existed in the clinical findings between the odontogenic and desmoplastic fibroma groups. After analysis of the data obtained from the literature, we then re-evaluated our three cases and attempted to determine if there was an odontogenic or nonodontogenic origin, i.e., whether the cases were intraosseous odontogenic or desmoplastic fibromas.

Case 1

Case Presentation

A 35-year-old male was originally seen at the hospital affiliated with Matsudo Dental College on October 7, 1990, and found to be suffering from swelling and pain in the right buccal region and was unable to fully open his mouth. A panoramic radiograph (Fig. 1) from the right mandibular premolar region to the ramus area showed an irregular margin of multilocular radiolucency and expansion of the lower border of the mandible. The lamina dura periphery 44 was not visible in the dental radiograph (Fig. 2). The CT image (Fig. 3) showed remarkable bone expansion from the mandibular premolar region to the ramus, along with a somewhat unclear and irregular margin. The mandibular canal ran through the lesion. The inner density was equal to muscle, and there were numerous small-radiopacified bodies visible.

Under general anesthesia, the patient underwent surgery on November 13, 1990. The excised specimen measured $8.2 \text{ cm} \times 2.5 \text{ cm} \times 1.3 \text{ cm}$, and the cut surface appeared grayish-white in color. The specimen consisted of proliferating fibroblastic spindle cells with oval or spindle nuclei. Collagen bundles were observed intermingled with the fibroblastic cells (Fig. 4). A diagnosis of intra-osseous fibroma was made.

Case 2

A 13-year-old female began reporting toothache at 36. She was referred by the practicing physician at the hospital associated with Matsudo Dental College to our dental hospital on March 2, 2000.

A panoramic radiograph (Fig. 5) showed a well-defined radiolucent area at the root apex of 35 and 36, demonstrating osteosclerosis around the periphery of the region. The mandibular canal was dislocated downward, and lamina dura periphery 36 was not visible in the dental radiograph (Fig. 6).

Axial CT scans (Fig. 7) showed a low-density area under the muscle, including the root apex of 36. Other findings in the CT image included thinning of the cortical bone and peripheral osteosclerosis. The mandibular canal was found to be dislocated downward as well.

Under local anesthesia, the patient underwent surgery on March 23, 2000. Several portions of the specimen,



Fig. 1 Panoramic radiography.



Fig. 2 Dental radiography.

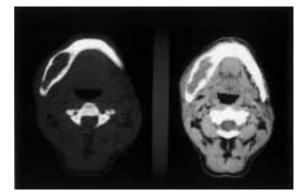


Fig. 3 CT image (Axial scan).

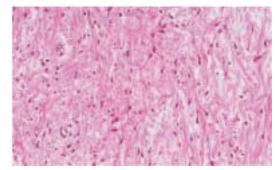


Fig. 4 Hematoxylin-Eosin stain (× 200).

measuring up to $0.5 \text{ cm} \times 0.4 \text{ cm} \times 0.4 \text{ cm}$, were extirpated. Proliferating, fibroblastic spindle cells with collagen bundles were evident, and hyalinization and overgrowth of the capillary vessels was observed (Fig. 8). Based on the data collected, a diagnosis of intra-osseous fibroma was made.

Case 3

A 51-year-old female, who reported pain and swelling in the left maxillary molar region, underwent a dental examination during an earlier visit with her personal care provider. The panoramic radiograph showed radiolucent area of 46. On June 21, 2001 she was referred by the practicing physician to Matsudo Dental College Hospital.

A panoramic radiograph (Fig. 9) taken at the hospital

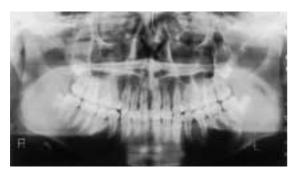


Fig. 5 Panoramic radiography.

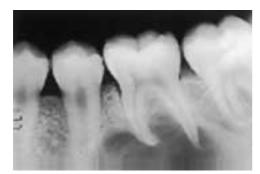


Fig. 6 Dental radiography.

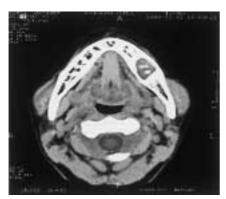


Fig. 7 CT image (Axial scan).

demonstrated entirely clear regions and a heart-shaped area of radiolucency that appeared to overlap the mandibular canal and mental foramen. An axial CT scan (Fig. 10) showed a low-density area under the muscle with an irregular margin, which included the apex of 46 and 47. The lesion reached the lingual cortical bone, and the mandibular canal ran through its center. There was swelling of the submandibular lymph node with a size of 2.0 cm × 1.0 cm.

Under general anesthesia, the patient underwent surgery on March 14, 2002. The excised specimen measured 1.7 $\text{cm} \times 1.5 \text{ cm} \times 0.7 \text{ cm}$, and the cut surface demonstrated a grayish-white color. The specimen consisted of proliferating fibroblastic spindle cells with oval or spindle nuclei and additionally, collagen bundles were observed

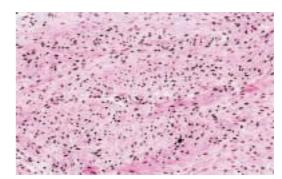


Fig. 8 Hematoxylin-Eosin stain (×200).



Fig 9 Panoramic radiography.

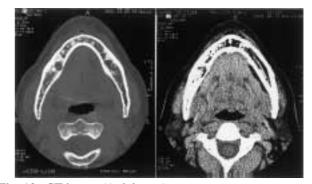


Fig. 10 CT image (Axial scan).

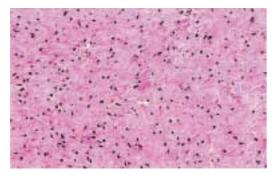


Fig. 11 Hematoxylin-Eosin stain (× 200).

 Table 1
 Comparison of odontogenic and desmoplastic fibroma (age and sex)

	Odontogenic fibroma	Desmoplastic fibroma
Age (No. of cases)	(64)	(68)
Mean	34.9 years	15.1 years
Standard deviation	19.6 years	12 years
Sex		
Male	19 cases	31 cases
Female	45 cases	38 cases

 Table 2
 Comparison of odontogenic and desmoplastic fibroma (location)

	Odontogenic fibroma	Desmoplastic fibroma
Maxilla (No. of cases)	(30)	(11)
Left	11 cases	3 cases
Right	17 cases	7 cases
Unclear	2 cases	1 case
Mandible (No. of cases)	(32)	(57)
Left	21 cases	28 cases
Right	10 cases	17 cases
Median	1 case	1 case
Unclear	0 case	11 cases

intermingled with the fibroblastic cells. Partial hyalinization and mucoid degeneration was also observed in the tumor (Fig. 11). A diagnosis of intra-osseous fibroma was made.

Case Analysis

To examine our hypothesis that intra-osseous fibroma can be classified as either central odontogenic or desmoplastic fibromas, we reviewed the literature and found 63 cases of intra-osseous (central) odontogenic fibroma reported from 1954 to 2002 and 69 cases of the desmoplastic fibroma reported from 1965 to 2001. The data from these cases are summarized in Tables 1-4.

Table 1 lists the age and gender distribution for both the intra-osseous odontogenic and desmoplastic fibroma groups and Table 2 indicates the location of occurrence of the intra-osseous odontogenic and desmoplastic fibromas. As indicated by the radiographic findings, Table
 Table 3 Comparison of odontogenic and desmoplastic fibroma (size)

	Odontogenic fibroma	Desmoplastic fibroma	
Under 1.0 cm	1 case	0 case	
Over 1.0 cm under 1.5 cm	1 case	2 cases	
Over 1.5 cm	19 cases	31 cases	

 Table 4 Comparison of odontogenic and desmoplastic fibroma (radiographic findings)

	Odontogenic fibroma	Desmoplastic fibroma
Shape		
Multilocular	15 cases	19 cases
Unilocular	12 cases	6 cases
Inner lesion		
Radiolucenct	48 cases	42 cases
Radiopaque	0 case	3 cases
Mixed	2 cases	1 case
Boundary		
Well-defined	9 cases	14 cases
Destructive	2 cases	8 cases
Bone expansion		
Presence	17 cases	22 cases
Root resorption		
Presence	7 cases	4 cases
Calcification		
Presence	3 cases	1 case

Table 5Differential diagnoses between central odontogenic
and desmoplastic fibroma (including our 3 cases)

	Central odontogenic fibroma	Desmoplastic fibroma	Case 1	Case 2	Case 3
Age	34.9±19.6y	15.1±12y	35y	13y	51y
Location	Max. and.	Almost	Mand.	Mand.	Mand.
	Mand	Mand.			
Destruction	3.1%	11.8%	Irregular	Well-	Well-
boundary	5.1%			defined	defined
Root resorption	10.9%	5.9%	NA	NA	NA
Calcification	4.7%	1.5%	NA	NA	NA

Max. = maxilla Mand. = mandibular

3 presents the dimensions of the intra-osseous odontogenic and desmoplastic fibromas, while Table 4 lists the other characteristics such as shape, inner lesion, boundary, bone expansion, resorption of adjacent tooth root, and whether or not there was enclosed calcification for both intraosseous odontogenic and desmoplastic fibroma cases.

There was only one report of intra-osseous odontogenic fibroma (29) that referred to CT images in contrast to ten reports for desmoplastic fibroma (51,54,60,63,65,67,69,72-74). Cases of desmoplastic fibroma were often observed

to involve the destruction or perforation of cortex bone. In the one report in which there was a MRI image of the desmoplastic fibroma, erosion of the right hard palate was observed (76).

Table 5 shows the differences between intra-osseous odontogenic and desmoplastic fibromas and includes the data from our three cases. Based on the differences we found in the reported cases in the literature, we were able to establish appropriate criteria for categorical guidelines. The differential diagnosis criteria were based on patient age, location of the lesion, boundary destruction, and calcification. For lesions that occurred in the maxilla, there was a high degree of probability that the correct diagnosis was intra-osseous odontogenic fibroma. However, in all three of our cases the lesions occurred in the mandible, which based on location alone suggested a diagnosis of either intra-osseous odontogenic or desmoplastic fibroma. Tooth root resorption was excluded as a criterion, since the differences observed from patient to patient were minimal. The resulting degree of resemblance calculations indicated that both cases 1 and 2 exhibited a 62.5% similarity to the desmoplastic fibroma cases, while case 3 exhibited a 62.5% similarity to the intra-osseous odontogenic fibroma cases. The major factor determining classification for case 2 was boundary destruction, which appears to be a decisive trait for the indication of desmoplastic fibroma.

Discussion

Fibromas of odontogenic origin are referred to as intraosseous odontogenic fibroma (1-3) while lesions of nonodontogenic origin are known as desmoplastic fibroma (1-3). Although odontogenic or non-odontogenic origin can be determined by the presence of odontogenic epithelium, in cases where there is an absence of odontogenic epithelium it is not possible to make similar clear-cut conclusions (77,78).

Kramer et al (77) have reported that odontogenic fibroma is a fibroblastic neoplasm that contains varying amounts of apparently inactive odontogenic epithelium. Safer et al (2) characterized such lesions as having the most poorly defined parameters. On the other hand, Regezi et al (3) described desmoplastic fibroma as being benign, locally aggressive lesions of the bone that share many features with soft tissue desmoid tumors and fibromatoses.

Three types of intra-osseous odontogenic fibromas have recently been proposed (1-3). The first type, (simple type), is a lesion around the crown of an unerupted tooth resembling a small dentigerous cyst. The second type, (WHO type), is a lesion described by the World Health Organization as a fibroblastic neoplasm with varying amounts of odontogenic epithelium and calcified material resembling dysplastic dentin or cement-like material. The third type, (granular cell type) (1), is a lesion that exhibits variable numbers of cells with an acidophilic granular cytoplasm. These three types are the recurring types that are encountered in the literature (13,31).

Desmoplastic fibroma is a locally aggressive lytic benign tumor of bone (1,53), with numerous cases involving children (34,38,43,45,57,59,60,70,73-75). Five such cases were found in the literature (56,62,68,79).

In the three cases from our institution, the apparent absence of odontogenic epithelium led to an original diagnosis of intra-osseous fibroma in all of the patients. Generally, such diagnoses are determined entirely by the presence of odontogenic epithelium. In other words, if odontogenic epithelium is present, a diagnosis of intraosseous odontogenic fibroma is made, while if it is absent there is a diagnosis of desmoplastic fibroma. As odontogenic epithelium was not found in all three of our cases, a diagnosis of intra-osseous fibroma was summarily made. Using the approach previously outlined, we set out to find another way of determining a diagnosis of intraosseous odontogenic fibroma or desmoplastic fibroma. Therefore we re-evaluated our three cases based on diagnostic data criteria that were determined from our analysis of the previously reported cases in the literature.

Desmoplastic fibroma tended to involve younger patients than that seen for intra-osseous odontogenic fibroma cases. The mean age for cases of desmoplastic fibroma was 15.1 years, while that for cases of intra-osseous odontogenic fibroma was 34.9 years (Table 1). Cases of intra-osseous odontogenic fibroma involved twice as many women as men, while cases of desmoplastic fibroma showed nearly equal sex ratios (Table 1). The cases of intra-osseous odontogenic fibroma occurred at locations nearly equidistant to the maxilla and mandible. On the other hand, desmoplastic fibroma occurred almost exclusively in the mandible (Table 2). The dimensions of both intraosseous odontogenic and desmoplastic fibromas were generally over 1.5 cm in diameter (Table 3).

Radiographic findings showed varying characteristics for points such as shape, inner lesion, boundary, bone expansion, and root resorption. Cases of intra-osseous odontogenic fibroma were nearly evenly divided between multilocular and unilocular patterns. On the other hand, cases of desmoplastic fibroma were three times more likely to show a multilocular than unilocular pattern. The inner portion of both lesion types tended to be radiolucent. The well-defined boundary of intra-osseous odontogenic fibroma was 4.5 times that of uncleared boundaries. The ratio for desmoplastic fibroma was just under 2 and the cases of desmoplastic fibroma were more likely to involve bone expansion than cases of intra-osseous odontogenic fibroma. Cases of intra-osseous odontogenic fibroma were more likely to involve adjacent root resorption than cases of desmoplastic fibroma (Table 4).

The surgical techniques that are applied are different between intra-osseous odontogenic and desmoplastic fibromas, as desmoplastic fibroma is more aggressive than intra-osseous odontogenic fibroma. An ostectomy is normally performed for intra-osseous odontogenic fibroma, while fundamental resection is done for desmoplastic fibroma (80).

With respect to the histo-pathological findings, intraosseous odontogenic fibroma has classically been divided into two distinct variants (1). The first variant is referred to as the simple type and resembles the dental follicle. It consists of a very bland connective tissue mass, and therefore can be described due to the many plump fibroblasts appearing equidistant from each other. Such lesions feature a few small islands of odontogenic epithelium scattered throughout the lesion. The second variant is designated as the WHO type and contains mineralized material variously interpreted as osteoid, cementum-like, or dysplastic dentin. Dysplastic dentin is usually found close to the odontogenic epithelium. The WHO type also features more islands of odontogenic epithelium (1-3). Recently a third type has been proposed and can be described as a granular cell variant of intraosseous odontogenic fibroma (1). These types of lesions are known as granular cell ameloblastic fibromas, central granular cell tumors of the jaw, granular cell odontogenic fibromas, and spongiocytic adamantinomas(1,28). In contrast, desmoplastic fibromas consist of interlaced bundles and whorled aggregates of densely collagenous tissue containing uniform spindled and elongated fibroblasts. Some areas may exhibit hypercellularity with plumper fibroblast nuclei. However, cytologic atypia and mitotic figures are not found and bone is not produced by this lesional tissue(1,3,47).

In conclusion, the findings of this study indicate an apparent significant difference between the factors found in subjects with intra-osseous odontogenic and desmoplastic fibromas. An analysis of data from previously published reports on 64 cases of intra-osseous odontogenic fibroma and 68 cases of desmoplastic fibroma found that factors such as age and gender distributions, location, and radiographic findings (boundary, root resorption, and calcification) were indicative of the specific type of fibroma. Based on this literature analysis, we re-evaluated the data from the three patients at our institution that were initially diagnosed as having intra-osseous fibroma. This reexamination resulted in cases 1 and 2 being classified as desmoplastic fibroma, and case 3 as intra-osseous odontogenic fibroma.

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