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CENTRAL GIANT CELL GRANULOMA AND CEMENTIFYING FIBROMA OCCURRING IN THE SAME LESION OF RIGHT MANDIBULAR BODY : A CASE REPORT

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A giant cell granuloma and a cementifying fibroma occurring in the same lesion of right mandibular body in a 10 year old boy is presented with a 12 month follow up without recurrence or any other complications after operation.

The relatively small lesion of cementifying fibroma was well delineated from the larger lesion of central giant cell granuloma, and as their origins are different each other(odontogenic or connective tissue origin), we have considered that both lesions had developed independently.

Clinically, the evidencesa of aggressiveness of giant cell granuloma were also found, that is, large size, earlier age of 10, root resorption of lower right lst annd 2nd molars and cortical perforation.

With curettage and electric cauterization, we have treated both lesions satisfactorily.

I. INTRODUCTION

The central giant cell granuloma of the jaws is a relatively uncommon pathologic process, accounting for 6.6% of all benign jaw tumors in one report of Austin et al., 1959.[1,2]

According to the first description of this lesion by Jaffe(1953)[3], central giant cell reparative granuloma was considered to be a local reparative reaction of bone to injury. But, in recent years, the word "reparative" has been deleted, since it is realized that many of these lesions are more "destructive" than "reparative"[4].

Clinically this lesion occurs most commonly in children or in young adults and has a predilection for females (F : M=2 : 1) and for mandible. The clinical behavior of central giant cell granuloma is variable. It ranges from a slow-growing asymptomatic lesion to an aggresive lesion that manifests itself with pain, root resorption, and a tendency to recur after excision.[5]

The radiologic finding of central giant cell granuloma is various except the characteristic of radiolucency, and many authors described "It is rather non-specific."[6]

On the other hand, cementifying fibroma composing about 2% of the odontogenic tumors presents as a solitary, well circumscribed radiolucent-radiopaque lesion. This occurs more commonly in young and middle-aged adults and shows marked predilection for occurrence in females(F : M=2 : 1) and in mandible.[7] This lesion is usually asymptomatic and displacement of teeth may be an early clinical feature.

Both the central giant cell granuloma and cementifying fibroma usually respond well to conservative treatment and the recurrence is rare.[8]

In our case, a central giant cell granuloma and a cementifying fibroma involved the same lesion in right mandibular body area. Although both lesions were reported by many authors respectively, we could not find the case similar to ours in the literature. And so, we reported this rare case with review of literatures.

II. CASE REPORT

A 10-year-old boy came to the Department of Dentistry, Inha General Hospital because of painless gingival

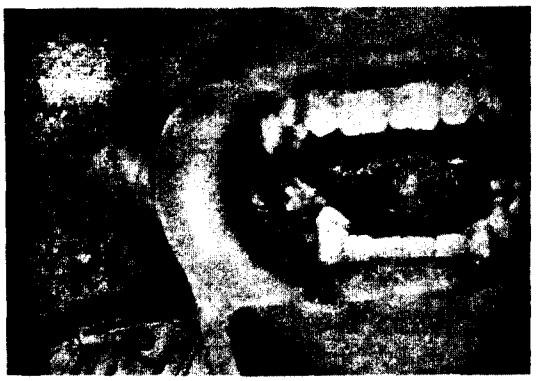

Fig. 1. Intraoral pre-op photo

Fig. 2. Pre-op panoramic view

overgrowth on the right mandibular lst molar area on December, 1989. The patient said that he did not feel any discomfort due to the mass.

On the physical examination, palpation of the head and neck revealed neither lymphadenopathy nor any other extraoral abnormality. Intraorally, firm and reddish gingival mass was located on the right lower 1st molar area, and the body of mandible was mildly expanded to buccal side(Fig. 1). The right lower 1st molar was partially embedded in the mass with mild mobility and positive response to percussion test. The lower 2nd premolar was missing state and the 1st premolar showed mild mobility and negative response to electric pulp vitality test.

In the radiographic examination, the panoramic view showed unilocular radiolucency in the apical region of right lower 1st and 2nd molars with resorption of their roots, and the inerior border of mandible was thinned. Another small lesion whuch was well defined by radiopaque border was noticed around the impacted 2nd premolar and supernumerary tooth which was thought to be unerupted 2nd deciduous molar, and it showed more increased density than the larger lesion(Fig. 2).

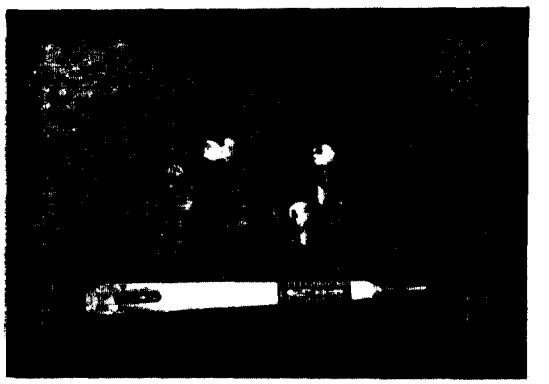

Fig. 3. Photo of the specimen

Under the tentative diagnosis of benign tumor of right mandibular body, we performed biopsy at the anterolateral periphery of the mass, and the result was giant cell granuloma.

The medical and familial history of the patient was not remarkable, and the laboratory data were within normal limit.

Five days later, the patient was taken to operating room and under general anesthesia, the 1st premolar was extracted. Then, an mucoperiosteal flap was was developed, and, after the lesion was isolated, careful curettage and dental extraction of 765 including supernumerary tooth were performed(Fig. 3). The bed was cauterized electrically and vaseline gauze was packed into the lesion. With daily dressing and nitro-furazone gauze change, the operation wound was healed without difficulty.

The most recent intraoral photo, 12 moths postoperatively, showed good healing state of the lesion without complications(Fig. 4).

Histologically, multinuclear giant cells were sparse and unevenly distributed in the stroma composed of small spindle shaped cells. The size of giant cells and the number of the nuclei were various(Fig. 5). In other portion, old hemorrhagic zone, resultant hemosiderin deposits, and infiltration of inflammatory cells were obser-

Fig. 4. Intraoral post-op(12 Months) photo

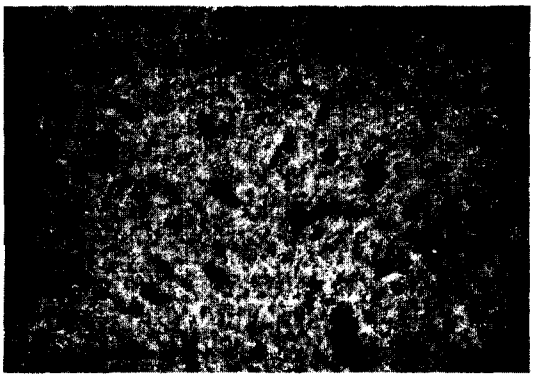

Fig. 5. Photomicrograph showing numerous giant cells(H&E, ×100)

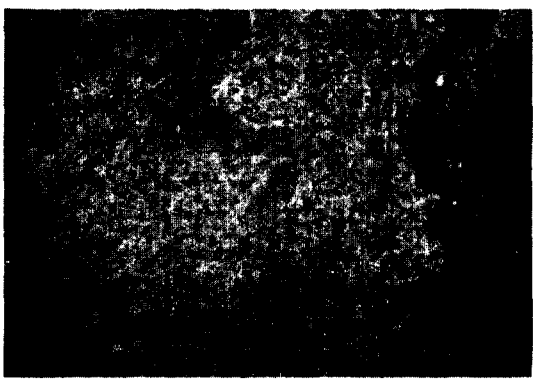

Fig. 6. Photomicrograph showing old hemorrhagic zone(H&E, ×100)

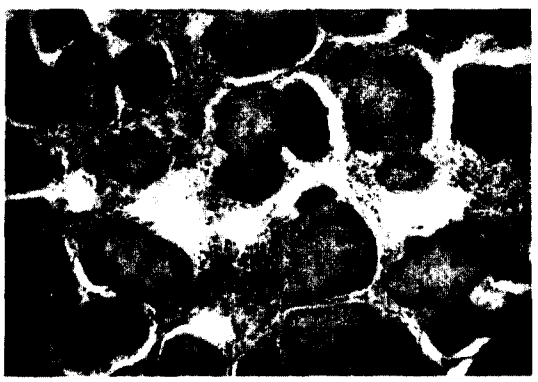

Fig. 7. Photomicrograph of various basophilic masses(H&E, ×100)

ved(Fig. 6).

The specimen taken from the premolar area, however, showed many basophilic masses which vary in shape and size, and the laminae of the mass formed concentric circles. At the periphery of the masses, eosinophilic zone and cementoblasts were observed(Fig. 7).

III. DISCUSSION

The giant cell granuloma that involved bone can be divided into two types-central and peripheral.[1,4,8,9] According to Austin et al.(1959),[1] central type is that in which a bony covering can be demonstrated either by roentgenographic examination or at the time of surgical exposure, and peripheral type occurs as alveolar epulides associated with varying degrees of underlying osseous resorption. And he considers that there appears to be little or no essential difference in the histology or the basic biologic behavior of both types. In 1973, Andersen and his associates[8] reported a clinical and histologic study of 129 cases of peripheral and central giant cell granulomas, and they concluded that both types of lesions were manifestations of the same pathological condition and division might be valueless.[10]

The giant cell granuloma of our case was regarded as central type which had perforated the cortical plate and grown into the oral cavity at the disto-buccal side of the right lower 1st molar region.

Central giant cell granuloma occurs in the jaws of young people, principally before the age of 30. It is more common in females, and the mandible predominates over the maxilla as the main site of occurrence. Pain is not

a prominent feature of this lesion, although some local discomfort is usually noted. And slight to moderate bulging of the jaw due to expansion of the cortical plates occurs in the involved area, depending upon the extent of bone involvement.

This lesion has a tendency to form in the anterior region of 1st molar in the mandible, frequently crossing the midline.[8, 11] About the site distribution, however, Cohen et al.(1988)[2] reported 31% of the lesions extended into the ascending ramus of the mandible, with two lesions involving the condylar process. And Horner(1989)[12] reported one case developed in the ramus of mandible beyond the tooth bearing region among the 26 cases of central giant cell granuloma and believed that central giant cell granuloma could not be excluded from the differential diagnosis of lesions posterior to the first molars.[13, 14]

The lesion of central giant cell granuloma of our case was located in the right lower 1st and 2nd molar area.

The etiology of central giant cell granuloma is extensively debated and remains uncertain. Many authors have followed Jaffe in considering the giant cell granuloma to be a local reparative reaction, possibly to intramedullary hemorrhage or trauma.[3, 15] Batsakis [7]proposed this lesion to be the result of injury imposed upon the periodontal membrane, the odontogenic mesenchyme, or the dental sac or its arcestral cells. In addition to these, several case reports have implicated hormonal activation by estrogen in the development of giant cell granuloma. Flaggert and associates(1987)[16] reported a case of recurrent giant cell granuloma occurring in the mandible of a patient on high dose estrogen therapy for the treatment of Soto's syndrome. Littler[17] and McGowen[18] also reported a relationship between the development of this lesion and pregnancy.

Radiographically, Jaffe[3] described the radiologic appearances as "rather non-descript". Austin et al.(1959)[1] who reviewed 24 cases, stated that most appeared as sharply delineated unilocular radiolucencies, whereas Waldron and Shafer(1966)[11] found the multilocular or soapbubble appearance to be characteristic. In 1988, Cohen and Hertzanu,[2] who analysed 16 cases, reported that the radiological features varied from an ill-defined destructive to a well-defined multilocular appearance. Recently, Horner (1989)[12] reviewed 26 cases of central giant cell granuloma and found that round or ovoid, unilocular, well defined but non-corticated radiolucencies were the most common feature. The overall radiographic picture of this lesion is not particularly specific.[6]

In the radiograph of our case, two lesions were revealed. One was well defined unilocular radiolucent lesion in the apical area of right lower 1st and 2nd molars which was diagnosed as central giant cell granuloma, and another, cementifying fibroma, showed radiopacity in the premolar area with well demarcated border.

The histologic pattern is a rather characteristic one. In a fairly loose, vascular stroma composed of small, spindle shaped cells, one notes a good deal of hemorrhagic extravasation. The multinuclear giant cells present are sparse, small, unevenly distributed, and often clumped in areas of hemorrhage. Some delicate trabeculae of newly formed osteoid or bone may also present.[3, 8, 15] The basic proliferating cells in all giant cell granulomas contain an oval or spindle shaped nucleus and have a prominent nuclear membrane and small nucleus.[1]

In 1986, Chuong and his associates[5] studied central giant cell lesions to search for possible histologic predictors of biologic behavior and to establish criteria for the diagnosis of the aggressive variant. According to them, the increased mean surface area occupied by giant cells and the increased mean relative size index of giant cells in clinically aggressive lesions revealed increased recurrences, and they concluded that aggressive giant cell lesions usually occurred in a younger age group, were larger at the time of diagnosis, and recurred more frequently(Table 1). And one year later, Ficarra et al.[19] reported a clinicopathologic and cytometric study of central giant cell lesions of the jaws and supported the results of Choung et al.

Judging from the clinical behaviors of the earlier age of 10, the relatively larger size, root resorption of right lower 1st and 2nd molars, and cortical perforation, we considered our case to be an aggressive lesion.

Lesions with histologic findings similar to those of

central giant cell granuloma of the jaws include hyperparathyroidism, cherubism, aneurysmal bone cyst, and true giant cell tumor of bone.[3,7,20,21,22]

Table 1. Giant Cell Lesions of The Jaw : Clinical Criteria

	Nonaggressive	Aggressive
Pain	no	yes
Rate of growth	slow	rapid
Swelling	variable	large
Root resorption	no	often present
Cortical perforation	no	often present
Recurrence	no	yes

Giant cell granuloma of the jaws had been interpreted simply as giant cell tumors and were thought to be similar to the true neoplastic giant cell tumors of long bones until Jaffe(1953)[3] who had found that there are a number of histologic and clinical difference between two lesions.[7](Table 2)[23] He had seen 1 example of a genuine giant cell tumor of the jaws, and Austin and associates(1959)[1] found 2 examples among the 66 giant cell lesions. And according to Shklar and Meyer(1961)[24], true giant cell tumors occur in the jaws with greater frequency than is believed by most authors, and they stated their experience with 10 cases regarded as examples of such lesions. In 1966, Waldron and Shafer[11] summarized the histologic criteria generally presented for diagnosis of benign giant cell tumors : (1) a moderately vascular stroma composed of plump spindle or oval shaped cells, with their nuclei surrounded by poorly defined cytoplasmic zones, (2) lack of significant production of intercellular substance, such as collagen or osteoid, by the stromal cells, (3) mitoses in the stromal cells, not uncommon and often quite numerous, and (4) a large number of giant cells scattered more or less evenly throughout the lesion.

The central giant cell granuloma of the jaws presents a very benign course and can be treated satisfactorily by enucleation or curettage. The frequency of recurrence may be in the range of 13%[8] but the recurrences can be treated by similar measures. Giant cell tumor of long bone, however, is locally aggressive with a high recurrence rate and the maligant transformation occurs in 15%

Table 2. Major Differences between GCG and GCT

	Giant cell reparative granuloma	Giant cell tumor
Age :	1st and 2nd decade	3rd and 4th decades
Location :	Mandible and maxilla most common, rarely occurring in the ethmoid, sphenoid, and temporal bone	Metaphysis and epiphysis of long bone. Uncommonly occurring in skull
Radiological study :	Lytic with erosion, but less likely to expand or perforate	Lytic with erosion : can become massive with extension into surrounding soft tissue
Histology :	Hemorrhage and hemosiderin, firbous stroma.	Diffuse, closely spaced giant cells
	Giant cells focally arranged and not numerous. Osseous formation	Uniform pattern
Radiation therapy :	Sarcomatous transformation has been reported	Relatively resistant, may be used as an adjunct. Sarcomatous transformation has been reproted
Course :	Benign. Heals by sclerosis and new bone formation. Recur only in 10~15%, even with inadequate removal	Aggressive with 45% recurrence rate

to 30% of the cases.[25] But the aggressive giant cell lesions of the jaws do not have high rate of malignant transformation.[5] Radiotherapy can also effect a cure of giant cell granuloma, but the possibility of postradiation sarcoma, though small, does exist.[15,25,26]

In our case, as the lesion was thought to be an aggressive type, additional electric cauterization was carried out after careful enucleation and curettage.

The cementifying fibroma composing about 2% of the odontogenic tumors presents as a solitary, well-circumscribed radiolucent-radiopaque lesion that usually occurs in the premolar-molar area of the mandible with marked predilection for females by a ratio of about 2 : 1. This lesion may occur at any age, but it is more common in young and middle-aged adults(mean age : 35 yrs). The lesion is generally asymptomatic until the growth produces a noticeable swelling and mild deformity, and displacement of teeth may be an early clinical feature.

Radiographically, the cementifying fibroma is well delineated and may appear as a radiolucency or a dense radiopacity, depending upon the amount of cementum within the lesion. As it shows a centrifugal growth pattern rather than a linear one, the lesion grows by expansion equally in all directions and presents as a round tumor mass.

The lesion consists of a cellular fibrous stroma containing scattered droplets of acellular cementum or rounded and fused basophilic masses of cementum like tissue. These islands are generally irregularly round, ovoid or slightly elongated, often lobulated.

The cementifying fibroma rarely recurs following conservative enucleation.[27,28]

In our case, the small and radiopaque lesion around the impacted teeth was relatively well defined from the main lesion of cemtral giant cell granuloma and diagnosed as cementifying fibroma finally.

Ⅳ. SUMMARY

This is a case report of central giant cell granuloma and cementifying fibroma involving the same lesion of right mandibular body.

At the time of operation, the relatively small lesion of cementifying fibroma was well delineated from the larger lesion of central giant cell granuloma. And as the former is odontogenic origin, whereas the latter is connective tissue origin, it is considered that both lesions were occurred independently.

In the clinical point of view, our case seems to be an aggressive giant cell lesion because of the relatively larger size occurring in the earlier age of 10, root resorption of 1st and 2nd molars, and cortical perforation.

With curettage and electric cauterization, we have treated both lesions satisfactorily, but, since the nature of aggressiveness of central giant cell granuloma is suspected, periodic recall and check-up will be necessary.

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국문초록

하악 우측 골체부에서, 동일한 병소 내에 발생한 골내성 거대세포 육아종과 백아질 섬유종의 치험례

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저자등은, 치은부의 종괴를 주소로 내원한 10세 남아의 하악 우측 골체부에서, 동일한 부위에 함께 발생한 골내성 거대세포 육아종과 백아질 섬유종으로 진단된 증례로, 수술시 비교적 작은 크기의 백아질 섬유종은 골내성 거대세포 육아종과 비교적 경계가 잘 지워져 있었고, 두 병소의 발생기원이 서로 다른 점으로 미루어 이들 두 병소는 서로 독립하여 동일 부위에 발생한 것으로 사료되며, 임상적인 관점에서, 거대세포 육아종은 어린나이에 비교적 병소가 크고, 제1 · 제2 대구치의 치근 흡수 및 피판의 천공 소견을 보여 aggressive type으로 판단되어, 소파술과 전기 소작술을 이용하여 두 병소를 만족스럽게 치험하고, 1년이 지난 현재까지 수술에 따른 후유증이나 재발의 소견을 나타내지 않기에 문헌고찰과 함께 보고하는 바이다.