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A CASE REPORT AND RIVIEW OF LITERATURE ON OSSIFYING FIBROMA IN MAXILLA

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Abstract

Ossifying fibroma is a relatively slow growing tumor, and likely to have presented for some years before its clinical diognosis. The usually well circumscribed nature of ossifying fibroma in jaws lends itself to relative ease of excision and hence the favorable therapeutic results. On occasion, however, particulary in juvenile patient, if maxilla the tumor assumes an aggressive behavior.

In that case, because the tumor grows invasively, resection with a margin of healthy tissue is indicated. The case presented is 34-year old female. The patient had noticed a gradual swelling of the right side of the face approximately 2 months in duration correlating with a intermittent pain on the right maxillary molar area. Palpation disclosed firm swelling on the right anterior and lateral walls of the maxillary sinus extended to the maxillary tuberosity area.

The radiographic examination revealed soft tissue mass with multiple dense round calcifications with destruction of anterior and posterolateral wall of the right maxillary sinus and right alveolar process, and hard palate. The mass totally obliterated maxillary sinus and extended to the pterygopalatine fossa.

The histologic diagnosis from the biopsied specimen revealed ossifying fibroma. The tumor mass was resected by subtotal maxillectomy procedure due to a recent rapid infiltrative growth. In 5 months of postoperative follow-up period, the patient has favorable prognosis.

INTRODUCTION

The ossifying fibroma is a benign, relatively slow-growing lesion in which the osteoblasts proliferate and forming a new bone, and is one of the fibroosseous lesions[1].

The pathogenesis of the ossifying fibroma is unclear[2] and there is no evidence to associate this lesion with other systemic conditions[3].

The primary clinical findings of the ossifying fibroma are firm lacalized swelling, disfigurement, and malocclusion[4]. The most important type of this group is the aggressive large lesion that usually producing progressive swelling and facial asymmetry[5].

The radiographic appearance is extremely variable, depending upon the stage of development, and ranges from a radiolucent lesion with haphazardly arranged calcification within the tumor mass to a dense radiopaque mass that well demarcated from the normal bone[6].

Histologically, the ossifying fibroma is composed of interlacing collagen bundles, proliferating fibroblasts, and irregular bony trabeculae[2].

Regardless of the character of the hard tissue component of the tumor, enucleation or conservative excision remain to be the preferred treatments[5].

Recurrence is rare, unless the primay removal was improper.

In juvenile patient if the tumor is in maxilla, it assumes an aggressive behavior. In that case, because the tumor grows invasively, resection with a margin of healthy tissue is indicated.

This case presented is ossifying fibroma which was developed on the maxilla in 34-year-old female.

CASE REPORT

A 34-year-old female presented with a gingival mass on right maxillary molar area, presented in Department of Oral & Maxillofacial Surgery, Kyung Hee Dental Hospital, Seoul, Korea, on March 13, 1989.

She stated that the lesion had been presented for 2 months, and right maxillary second molar had extracted about a year ago at the local dental clinic due to pain. She has complained the persisted pain on right maxillary molar area. We observed firm mass on right maxillary molar area, tenderness to palpation, and facial asymmetry on the side of the lesion. During 2 months prior to visit, she stated that tumor was growing rapidly (Fig. 1).

Fig.1. A photography on the lesion.
The lesion on the right maxillary molar area is firm mass, tenderness to palpation, and extended to the tuberosity area.

The radiographic examination on this lesion was performed through panoramic, Waters', posteroanterior, occlusal views, and periapical dental radiographs, and computed tomography. A radiolucent mass containing calcified material was present in the right maxilla, extended into the right maxillary sinus, lateral wall of nasal cavity, anterior border of right maxilla, and inferior floor of right orbit. Panoramic and periapical dental radiographs, as well as computed tomograph, suggested calcification within the soft tissue mass. Regular and well-defined borders were presented on the periphery of the lesion. But in the anterior part of the lesion, the border was ill-defined and the invasive bony destruction was observed on the apical area of premolar(Fig. 2).

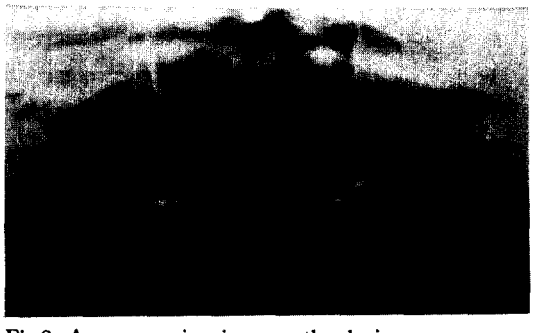


Fig.2. A panoramic view on the lesion.
Regular and well-defined borders presented at the periphery of the lesion. But in the anterior part of the lesion, the border is ill-difined and invasive bony destruction is reveales.

In the periapical dental radiographs, the anterior margin of the lesion is ill-defined and infiltrative to the adjacent bony tissue on the right maxillary canine (Fig. 3).

On the computed tomographic facial scan, the relatively well marginated soft tissue mass with multiple calcifications and destruction of the posterolateral wall of the right maxillary sinus and alveolar process of the maxilla were noticed. But the pterygoid plate and medial nasal wall were intact. On coronal scan,

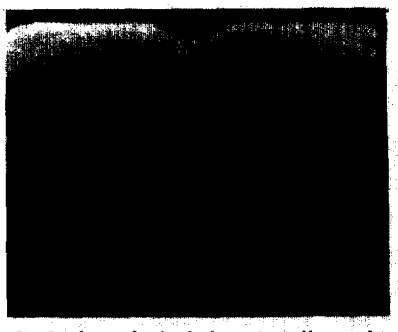
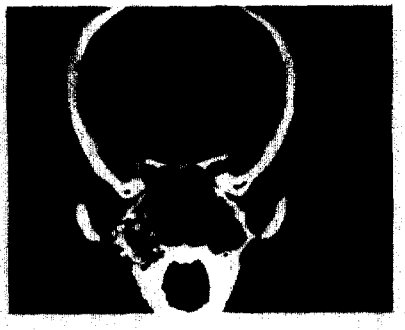


Fig.3. A periapical dental radiographs on the lesion. The anterior margin of the lesion is illdefined and infiltrative to the adjucent bony tissue on the right maxillary premolar area.



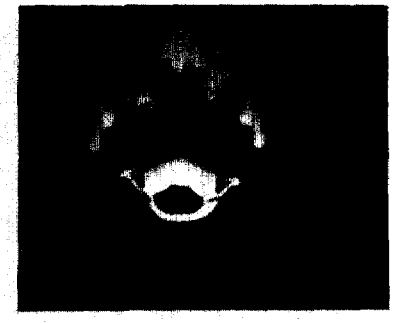


Fig.4. Computed tomography on the lesion. The relatively well marginated soft tissue mass with multiple calcifications and destruction of the posterolateral wall of the right maxillary sinus and alveolar process of the maxilla were noticed.

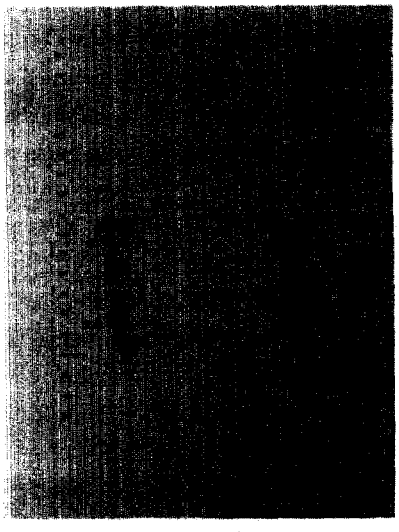


Fig.5. A Nuclear bone scan on the lesion. It shows intense activity at the right maxillary sinus and alveolar process of maxilla.

Fig.6. A light microscopic view on the lesion. The connective tissue is composed of exceedingly cellular mass comprising large numbers of plump proliferative fibroblasts, randomly scattered trabeculae of actively forming bone.

the destruction of the right hard palate is well delineated with multiple calcifications. And the mucoperiosteal thickening of the posterior wall of the left maxillary sinus is seen (Fig. 4).

Nuclear bone scan using 99 M technetium methylene diphosphonate (99m Tc MDP) showed intense increased activity at the right maxillary sinus and alveolar process of maxilla, which may be corresponding to radiographic bony lesion site (Fig. 5).

The light microscopy revealed a rubbery tissue which is composed of covering epithelium and connective tissue. The underlying connective tissue is composed of exceedingly cellular mass of connective tissue comprising large numbers of plump proliferative fibroblasts, randomly scattered trabeculae of actively forming bone. Occasionaly multinucleated giant cells were seen, but they are not numerous (Fig. 6).

On the ultrastructural study, tumor tissue is consi-

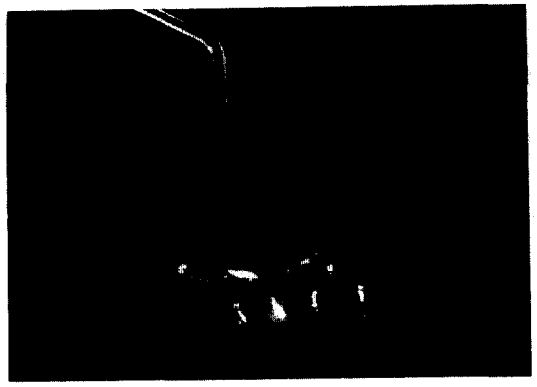


Fig.7. The excised specimen of the lesion.
It was a 10×7×5cm tumor mass which has removed via subtotal maxillectomy procedure.

Fig.8. A Partial denture-type resin obturator.
It is replaced intraorally at 3 weeks after operation, to compensate the phonetic and masticatory disturbances postoperatively.

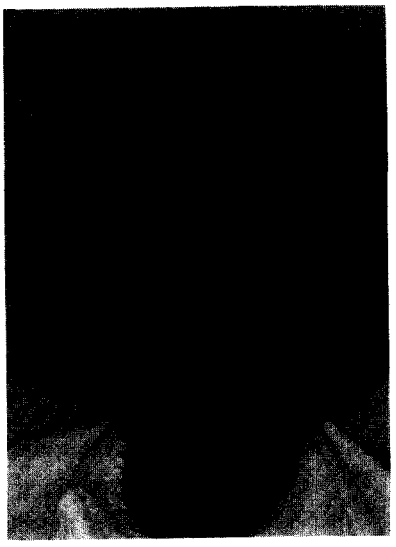


Fig.9. An anterior aspect of the patient a 5 months, postoperative follow-up.
It shows good postoperative esthetic results.

sted of proliferating fibroblasts with collagen formation.

In summation we diagnosed ossifying fibroma on the right maxilla. The patient was admitted on March 28 1989 and prepared for surgery.

The next day, with the patient under nasotracheal general anesthesia, the tumor was approached by Weber-Fergusson incision for the better access to the posterior wall of the maxilla. Ostectomy on anterior portion of maxilla was performed with a Stryker saw. And then ostectomy of infraorbital area and palatal area was performed. Finally, subtotal maxillectomy procedure was performed with osteotome, preserving the right infraorbital rim and medial wall of nasal mucosa (Fig. 7).

And then, partial thickness skin graft from the right thigh was obtained with dermatome, and skin graft was sutured on the defect intraorally, and then resin obturator was placed with 24G wire on upper left central incisor and first molar.

A 10×7×5cm tumor mass was excised, ossifying fibroma was diagnosed on post-operatire pathologic examination.

Following an operation, she complained phonetic disturbance. At 3 week after operation, the surgical defect was replaced by partial denture type obturator (Fig. 8), and the phonetic & masticatory functions are restored without remarkable problems. In 5 months post-operative follow up period the patient showed favorable prognosis (Fig. 9).

DISCUSSION

The ossifying fibroma is a benign, relatively slow-

growing lesion in which the osteoblasts proliferate and form new bone, and is one of the fibroosseous lesions[1]. Benign fibro - osseous lesions include ossifying fibroma, periapical cemental dysplasia, proliferative periostitis of Garre, focal sclerosing osteomyelitis, and osteitis deformans. These lesions share common microscopic features[7]. Therefore, ossifying fibroma of bone has caused considerable clinical controversy because of confusion with the terminology and the criteria for diagnosis[7]. But it is a benign neoplasm that recognized as of distinct pathologic entity, separated from most other fibro - osseous lesions those are not true neoplasms[2].

Since 1872, when Menzel[8] first described the entity now known as ossifying fibroma, there has been confusion over the lesion among surgeons, radiologists and pathologists. It was not until 1927 when Mongomery[9] used the term ossifying frbroma by which the lesions is now known. Since the introduction of the term "fibrous dysplasia of bone" by Lichtenstein[4] in 1983, there has been an increasing tendency to apply this term to a group of fibro - osseous growths frequently seen in the jaw bones, and it was accepted that the ossifying fibroma and fibrous dysplasia is one and the same, or one as a variant of the other. Lichtenstein and Jaffe[4] suggested that lesions diagnosed as ossifying fibroma actually represent a monostotic form of fibrous dysplasia. Cahn[10], Fisher[11], and Robinson[12] also have suggested that in most cases of previously reported ossifying fibroma are actually examples of fibrous dysplasia.

In 1946, Billings and Rigertz[13], while applying the term fibroosteoma, expressed doubts that these lesions are true neoplasms.

In 1950, Thoma[14] has evolved a calcification for these lesions based on the degree of differentiation in the various tissues. He designates these lesions as ossifying fibromas, fibro - osteoid - osteoma and mature fibroosteoma.

Since 1948, when an excellent review of the radiologic appearance of the ossifying fibroma was reported by Waldron and Giansanti[15], most investigators have considered the two lesions as separate and distinct clinical entities. And then, they have attempted to differentiate and identify these lesions with respect to the histologic presence or absence of cementum - like structures.

In 1968, Hamner and others[16], in a comprehensive analysis of 249 cases of fibro - osseous lesions of the jaws, suggested that the lesions are products of multipotential mesenchymal blast cells situated in the periodontal membrane that have the capacity to produce cementum, alveolar bone, and fibrous tissue. Therefore, they differentiated these lesions as cementoid, osteoid, mixed or fibrous, based first on the presence or absence of with hematoxylin and eosin and the degree of fineness of parallel birefringent lines seen with polarized light microscopy.

In 1976, Langdon and others[17] suggested that because there is no absolute histologic distinction between bone and cementum, and as cementum - like calcifications are seen in fibro - osseous lesions of all membrane bones, the distinction between ossifying and cementifying lesions should be discontinued. They also suggested that ossifying fibroma do not represent a single entity, but are a point on a spectrum of fibro - osseous lesions that included fibrous dysplasia as well.

The pathogenesis of the ossifying fibroma is unclear[2], Thoma[18] said that the lesion may develop from cell rests in normal bone. Waldron and Giansanti[15] on the other hand, believed that the lesion has a fibroblastic origin, and said that other investigators have presented evidence that the lesion arises from the periodontal ligament.

There is no evidence to associate this lesion with systemic conditions affecting calcification of bone such as Pagets disease or the brown tumor of hyperparathyroidism. Although the maxilla and mandible may be involved simultaneously, reports of a more extensive polyostotic involvement are lacking. The long bone may be affected, but no simultaneous association with oral lesions has been reported.

No hereditary predispositions are classically desc-

ribed for the central ossifying fibroma. However the odontogenic origin lesion related to the sclerotic cemental masses, described by Waldron and others, the predilection of sex, age, and race may be applicable, bacause the patient usually is in a middle - aged black woman. In our case, etiology of the lesion is unknown[2].

In 1985, by Eversole and others[7], sixty - four cases were classified as ossifying and/or cementifying fibroma on the basis of the following criteria; (1) clinical evidence of cortical expansion, (2) radiographically well - defined lesional borders, and (3) histopathologic features of a benign fibro - osseous process. The average age was reported to be 36 years, with a predilection for the third and fourth decades accounting for 56% of the sample. Fifteen percent occurred on persons under the age of 20, while the remaining patients exceeded 40 years of age. The female - to - male ratio was reported to be 5 : 1. Racially, 47% were white, 16% were black, 11% were Asian, 24% were Hispanic, and 2% were American Indians. The mandible was the site of predilection, accounting for 89% of the cases. Lesions arose most frequently in the molar region (52%), followed by the premolar area(25%), incisor area(13%), and cuspid region(11 %). On radiographic measurements they varied in size from lcm to more than 5cm in diameter. In our case, the patient is 34 - year - old female, and the lesion was occurred in the molar area, and these were agreed to predilection. But the lesion was occurred in maxilla, contrary to the predilection.

The primary clinical findings of the ossifying fibroma are firm localized swelling, disfigurement, and malocclusion[2]. The lesion is generally asymptomatic until growth produces noticeable swelling and asymmetry[6]. Because of the characteristic slow growth, the overlying mucosa and cortical plates of bone are usually intact unless injured[2]. The lesion is typically well circumscribed, compared to fibrous dysplasia, and the surrounding tissue is normal in appearance. Although multiple lesions may be seen in the maxilla and mandible, polyostotic involvement such as that seen in fibrous dysplasia does not occur[18]. Displacement of adjacent teeth is common, as well as impingement upon adjacent structures[6]. But there is usually no neurological disturbance[19].

The majority of fibro - osseous lesions of the jaws are small, single or multiple lesions that are noticed incidentally on routine radiographic examination[5]. Clinically, the most important members of this group are the aggressive large lesions which produce progressive swelling and facial asymmetry[5]. The aggressive ossifying fibroma appear most often in young patients, almost always younger than age of 40 and predominantly under age of 20, without racial or sexual predilection[5]. Incidence of these lesions is small enough for them to be considered rare; when they are present, there is difficulty in differentiating between these and fibrous dysplasia or osteosarcoma[5]. Although most reports characterize the lesion as slow growing over a period of years, but there can be a rapid increase in size in a relatively short period of time[5]. The mandible and maxilla appear to be equally affected, and there also have been reports of involvement both of the anterior base of the skull and of the temporal bone[20,21]. Large lesions may outgrow their supply of blood and become secondarily infected[2]. The patient may have neuro sensory disturbances that range from pain to numbness in the involved area. When the tumor is in maxilla, symptoms may include nasal stiffness and epiphora on the affected side. There also may be an associated exophthalmus and visual disturbances, depending on the extent of compression of orbital contents by the tumor[5].

Hamner and others[16] arbitrarily defined the larger lesions that involve the space occupied by two or more teeth, or being larger than 2×2cm, excluding true cementomas, as tumors. By these criteria, in their survey of 249 lesions, they found three in the maxilla that they termed cementoid lesions; seven that were termed osteoid (five in the mandible and three in the maxilla). Waldron and Giansanti[15], in their series of 43 lesions, found 13 that were seen

as tumors; five cementifying fibroma, six ossifying fibroma. In our case, the patient complained persisted pain on right maxillary molar area. And we observed firm mass on right maxillary molar area, tenderness to palpation, and facial asymmetry due to the lesion (Fig. 1).

The radiographic appearance is extremely variable, depending upon the stage of development, and ranges from a radiolucent lesion with haphazardly arranged calcification within the tumor mass to a dense radiopaque mass well demarcated from the normal bone[6].

Sherman and sternberg[20] drew several conclusions from the review of 12 cases that classified as giant lesions by the standards of Hamner and others were reported in an early radiographic review of the ossifying fibroma.

First, the lesion was unilocular, oval, or spherical, with a distinct osteolytic boundary delineating the tumor from normal bone. Second, as the lesion enlarged, there was a progressive increase in radiopacity, but it has less overall density than that of normal bone. Third, tooth displacement was reproted as well as instances of partial destruction of roots of teeth. Finally, maxillary tumors produced unique growth patterns in which there was dissolution of neighboring bones without displacement by pressure.

Radiographically, the ossifying fibroma is distinguished from fibrous dysplasia by the difference in radiodensity, architecture of the lesion, and the character of the borders of the lesion. Ossifying fibroma is somewhat radiolucent with haphazardly arranged calcification within the tumor mass. Fibrous dysplasia is often characterized by a radiopaque area with a ground - glass appearance. Regular and well - defined borders are present at the periphery of an ossifying fibroma, and expansion may disrupt the normal bony anatomy. The borders of fibrous dysplasia are indistinct and the advancing front of the lesion blends into the surrounding bone. Expansion of the host bone may also occur in fibrous dysplasia[20,22,23,24].

Resorption of roots is not common with fibrous dysplasia and other fibro - osseous lesions, but is often seen in ossifying fibroma[5].

In 1985, by report of Eversole[7], all neoplasms originate in tooth - bearing regions, and none were associated with the crowns of impacted teeth. By definition, all exhibited well - demarcated borders, or multilocular radiolucencies. Root divergence was featured in 17% of the instances, while root resorption was seen in 11%, and 35% were detected in edentulous areas.

In the maxillary lesions, a thin shell of bone usually is present along the outer border of the tumor.

Waldron and Giansanti[15] noticed in their study that the anterior maxilla appears to be spared from involvement.

In a series of 121 patients with tumors in the maxilla and mandible, all but one were radiolucent and the average size of the tumors was 5×4×ccm[15].

In our case, a radiographic mass containing calcification was present in the right maxilla, extended into the right maxillary sinus, lateral wall of nasal cavity, inferior floor of right orbit. On nuclear bone scan, intense increased activity was seen at the right maxillary sinus and alveolar process of maxilla. On computed tomography, multiple calcifications were noticed. The size of the lesion is about 10×7×5cm (Fig. 2, 3, 4, 5).

Ossifying fibroma elaborate bone, cementum, and spheroidal calcifications, prompting many pathologists to assign various terms to these benign fibro - osseous neoplasm. When bone predominates, ossifying fibroma is the appellation, while the term cementifying fibroma has been assigned when curvilinear trabecular or spheroidal calcifications are encountered. When bone and so - called cemental tissues are observed, the lesions have been referred to as cemento - ossifying fibroma[25].

Histologically, the ossifying fibroma is composed of interlacing collagen bundles, proliferating fibroblasts, and irregular bony trabeculae[2].

Since the mesenchymal progenitor cells of the periodontal ligament are capable of elaborating both bone and cementum. Hamner, Waldron and Giansanti

have surmised that ossifying and cementifying fibromas are benign fibro - osseous lesions of periodontal ligament origin and therefore represent histologic variations of the same neoplastic process[1, 15].

Some ossifying fibroma behave in an aggressive manner(?) reaching massive proportions with extensive cortical expansion. Most of these aggressive lesions occur in children and have promoted the designation juvenile aggressive or active ossifying fibroma [26].

Shafer and others[11] have suggested that ossifying and cementifying fibromas are either two distinct tumors developing on parallel lines, one arising from osteoblasts and the other arising from cementoblasts, or that they are facets of the same tumor. From the predictive standpoint, this differentiation holds little values, and the large aggressive lesions of either the ossifying or the cementifying type vary little with regard to age and sex of the patients, and locations or behavior[15, 17, 23].

The differentiation of fibrous dysplasia and aggressive ossifying fibroma is possible through histological examination[1]. Hamner and others[16] describe fibrous dysplasia as characteristically having a fibrous stroma with loose myxomatous areas, a good blood supply, and feathery, irregularly shaped trabeculae of woven bone, which lacks rimming by osteoblasts.

Fibrous dysplasia also shows random birefringence under polarized light, in contrast to the ossifying fibroma which has parallel dark and light birefringent lines.

As Small and Goodman[22] graphically demonstrated, if this is the best criterion, a biopsy specimen must contain an area of normal bone to be diagnostic. Pathologically, other criteria regarding internal structural differences have been identified to separate the two lesions. First, osteoclasts and osteoblasts tend to be more prominent in the ossifying fibroma.

Second, a comparatively more regular trabecular pattern is seen on ossifying fibroma. Third, ossifying fibroma has a stroma that, compared with fibrous dysplasia, contains fewer collagen and vascular elements and more cellular elements.

In 1985, Eversole and others[7] reported histologic features of 64 cases of ossifying and cementifying fibroma by means of both routine light microscopy and polarization microscopy. Four basic hard - tissue configurations were observed, including woven bone trabeculae, lamellar bone trabeculae, ovoid - curvoid deposits, and anastomosing curvilinear trabeculae. In woven trabeculae, polarization microscopy disclosed a meshwork of thick collagen fibers exhibiting a cross - hatched pattern. Lamellar trabeculae evinced parallel orientation of thick collagen fibers. One case exhibited a pagetoid mosaic pattern. Two types of ovoid, curvoid, or spherical calcifications were apparent. One type appeared dystrophic and acellular, with irregular margins. These dystrophic products were usually associated with a storiform pattern within the fibroblastic stroma. The second type was characterized by smooth boundaries evincing compacted spheroid - curvoid calcifications lying within a hypercellular fibroblastic stroma lacking a storiform orientation. The fourth pattern was characterized by smooth, often anastomosing, trabeculae with curvilinear configurations.

Under polarized light, the fiber width was fine and was arranged in a guilted network or demonstrated a microlamellar orientation. Dense cortical bone - like deposits were observed in some instances and were surrounded by more immature fibro - osseous elements. These patterns appeared most frequently in woven trabeculae(63%), followed by spheroid - curvoid deposits(44%), curvilinear trabeculae(41%), lamellar trabeculae(31%), and dense foci(30%).

In our case, underlying connective tissue is composed of exceedingly cellular mass of connective tissue comprising large numbers of plump proliferative fibroblasts, randomly scattered trabeculae of activity forming bone in light microscopy. Occasionary multinucleated giant cells were seen but they are not numerous (Fig.6).

Definitive treatment may be delayed because of the difficulty in establishing a correct diagnosis[5]. Re-

gardless of the character of the hard tissue component of the tumor, enucleation or conservative excision remain the preferred treatments. Because the lesion is separated and shelled out from the surrounding bone, surgical excision is not difficult[5]. In our case, because of its location in the alveolar region, the anterior margin of the tumor mass was invasive and the anterior wall of the maxillary sinus was destructed, we have removed tumor mass via subtotal maxillectomy procedure. Recurrence is said to be rare, unless incorrectly removed[5] in the initial approrch (Fig.7).

In report of Eversole[7], adequate follow-up data with radiographic documentation was obtained in twenty-three cases. The mean followup period was 38 months, with a range of 12 to 120 months. Thirteen resolved, whereas five recurred. Overall initial recurrence rate is 28%.

No significant correlations were found when radiologic and histologic features, including both product heterogeneity and stromal configuration, were evaluated in both resolved and recurrent cases.

Furthermore, aside from the perivascular hyalinization in one aggressive lesion alluded to previously, no microscopic differences were encountered with the large aggressive tumors, were compared with smaller neoplasms.

In our case, the patient complained phonetic disturbance and masticatory disturbance. At 3weeks after operation, partial denture type obturator was replaced, and these problems were resolved. At present, 5 months after operation, the patient has will adapted to the obturator and is remarkably comfortable with the device

CONCLUSION

A 34-old female with a gingival mass on right maxillary molar area, presented on the Department of Oral and maxillofacial surgery, Kyung Hee Dental Hospital, Seoul, Korea, on March 13. Through the clinical, radiological, and histological examination, we had diagnosed ossifying fibroma.

The tumor mass was removed totally via subtotal maxillectomy procedure in consideration of rapid growth & ill-defined margin of the mass. In follow up period of 5 months after operation, we have good result without remarkable functional problems or evidence of recurrence.

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상악골에 발생한 화골성 섬유종의 증례보고 및 문헌고찰

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국문초록

화골섬유종은 비교적 천천히 증식하는 종양으로 발견전 몇년간은 나타나지 않는다. 악골에서 화골섬유종의 잘 경계되어진 성질은 완전한 제거가 용이하여 이로인한 치료결과도 양호하다. 그러나 때때로 유년기 하악에서 이 종양의 골격적 양상을 볼 수도 있다. 그러한 경우 종양의 침윤적인 성장 때문에 건강한 조직을 포함한 절제가 적용된다. 본 증례의 환자는 34세의 여성으로 약 두달전 우측 안모의 점진적인 팽윤을 인지하였으며 상악우측 구치부의 간헐적인 동통을 호소하였고 종물은 상악결절부위 및 우측 상악동의 전측방벽까지 팽윤되어 있었다. 방사선적 조사에서 우측 상악동의 후측방벽과 전방벽 그리고 우측 치조돌기와 경구개 까지의 파괴와 함께 다수의 치밀한 구형의 석회화 물질을 함유한 종물이 관찰되었다.

종물은 최근의 빠른 침윤성 성장을 고려하여 부분적 상악골 절제술로 제거되었으며 6개월간 술후검사에서 환자는 특이한 술후 이상을 보이지 않았다.