Primary Giant Cell Tumor of Soft Tissue in the Mental Region

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Abstract: Giant cell tumors of soft tissue (GCT-ST) are considered to be the soft tissue analogue of giant cell tumors of bone. Although the majority of GCT-ST cases manifest histologically benign features, special consideration is required because GCT-ST can occasionally demonstrate extensive local invasion or distant metastases. Here, we present an extremely rare case of GCT-ST arising from subcutaneous mental soft tissue, rapidly growing over the mandibular gingiva and bone, and lacking overtly malignant features in clinical, radiological and cytological findings.

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Clinical Report

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Introduction

The giant cell tumor (GCT), as previously defined the nature of the entity by Jaffe et al.1, is generally benign neoplasm but has propensity for recurrence2-4), malignant transformation5,6) or distant metastasis7,8) because of its aggressive clinical behavior. As previously reported9, approximately 80-90 % cases of GCT have been occurred in the extremities, especially in the lower extremities10. Most cases of GCT are located in bony tissue and the incidence of GCT arise in the soft parts is rare compared to that of its bony counterparts. Primary GCT of soft tissue (GCT-ST) was originally demonstrated by Salm and Sissons in 19729, followed shortly by Guccion and Enzinger10). In the oral and maxillofacial region, not frequent site for GCT, most cases affect the mandible and maxilla11-12) and some cases located in the salivary gland13,14) or nasal cavity15 have been reported as primary soft tissue analogue in this region. Despite the majority of GCT in the oral and maxillofacial region are histologically comprised of an abundance of multinucleate giant cells and homogeneous population of round to polygonal mononuclear cells in the background13,14) with poor cellular atypia and mitotic figures, some of them have revealed a carcinomatous component15,16,17,18,22,24) or malignant transformation like those in the extremities. Here we report a case of 37-year old woman with a well-defined mental subcutaneous giant cell tumor rapidly growing over the mandibular gingiva and mandibular intraosseous tissue. According to the JHTBFF report, the GCT-ST arising from the soft tissue in the mental region has not previously been reported as far as the authors are aware.

Case report

A healthy 37-year-old woman presented to an outside institution with a history of several months of swelling and pain over her mental region. Although administration of antibiotics and the drainage using transverse incision in the mandibular anterior region of alveolus were performed under a presumptive diagnosis of chronic alveolar abscess, the clinical symptoms were remained to be unchanged and the patient was finally given a referral to our institution on July 2005 for further evaluation and treatment.

During her initial visit, there was a subcutaneous, firm nodule on the mental region with the skin flare (Fig. 1A). The lump was gradually progressive in size, but the patient was uncertain of the exact duration of the lesion or the rapidity of growth. Intraoral examination showed dome-shaped submucosal swelling at lower anterior vestibular region. The lesion was relatively well-demarcated, elastic-hard and covered with normal mucosa (Fig. 1B). Neurological examination revealed no definite sensory disturbance at mental region and there was no associated cervical lymphadenopathy. Also there was no preceding history of trauma at the mental region. The patient had no past medical history of note such as parathyroid tumor and general physical examination and laboratory findings such as serum calcium, phosphate, alkaline phosphatase were normal.

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Roentgenographic findings

The plain radiographic examination with panoramic radiograph and lateral cephalogram revealed no distinctive intraosseous lesion in the anterior region of mandible and erosion and sclerosis of the mental cortical bone with a soft-tissue thickening (Fig. 2A and B). For further examination, the enhanced magnetic resonance imaging (MRI) were performed and a large well defined hyperdense contrast enhancing lesion originating from the peripheral soft tissue of the mental region was identified. The lesion demonstrated homogeneous enhancement and hypodensity when compared to the fatty tissue (Fig. 2C). No calcifications were noted within the mass. Chest radiographs was normal.

Operation

Under the clinical diagnosis of benign tumor at mental region, biopsy of the subcutaneous lesion underwent and the specimen was interpreted as GCT. The patient was taken up for surgery with an intention of radical removal. Marginal resection of the tumor underwent through intraoral and submental approach on Sep. 7, 2005 (Fig. 3A). A subperiosteal dissection was performed except the region, where the tumor was adherent to the labial anterior lower cortical bone and complete removal of the intact lesion was performed by the osteotomy of the adjacent bone cortex. Also the digastic anterior muscle was seen to be infiltrated by the tumor and was partially excised. Gross estimate of tumor size was 35 x 30 x 15 mm and the tumor was firm and reddish brown (Fig. 3B). The excised specimen was serially sectioned and revealed to be a poorly delineated lesion. After the surgery, the patient was gradually relieved of pain and discharged from hospital two weeks later. Neurological deficit, a numbness on the lower lip to mental region, was present on discharge and gradually relieved several months later. Five year and five months after the
operation, the patient had no evidence of metastasis or recurrence from the inspection and follow-up MRI.

**Histology**

The lesion was fixed in 10% neutral formalin and stained with hematoxylin and eosin. Microscopically, the majority of the lesion was located in the soft tissue of the mental region, mostly covered by myo-fibrous tissue, but not completely encapsulated. It had a multi-nodular form in which the nodules were separated by fibrous septa of various thicknesses (Fig. 4A). These nodules contained a large number of osteoclast-like multinucleated giant cells diffusely distributed in the background, and also contained mononuclear stromal cells with round or oval nuclei, accompanied by small blood vessels (Fig. 4B). Neither multinucleated nor mononuclear cells exhibited distinctive cytological atypia (Fig. 4C). The lesion partially invaded cortical bone with reactive bone formation and bone resorption, but not into the digastric anterior muscle attached to the lesion (Fig. 4D-F). In addition, there was little extensive vascular / lymphatic invasion by tumor thrombi into small lesional vessels.
Table 1. Literature Review of Giant Cell Tumor of Soft tissue in the Oral and Maxillofacial region

<table>
<thead>
<tr>
<th>Authors (year)</th>
<th>No.</th>
<th>Cases</th>
<th>Age/Gender</th>
<th>Site</th>
<th>Treatment</th>
<th>Disease Outcome</th>
</tr>
</thead>
<tbody>
<tr>
<td>Eusebi et al (1984)</td>
<td>3</td>
<td>30/M, 43/M, 52/M</td>
<td>Parotid gland</td>
<td>Surgery</td>
<td>NED (4 years, 6 years, 4 years)</td>
<td></td>
</tr>
<tr>
<td>Balogh et al (1985)</td>
<td>1</td>
<td>67/M</td>
<td>Parotid gland</td>
<td>Surgery and RT</td>
<td>DWD (28 months after surgery)</td>
<td></td>
</tr>
<tr>
<td>Batsakis et al (1988)</td>
<td>2</td>
<td>59/M, 92/M</td>
<td>Parotid gland</td>
<td>Surgery</td>
<td>NED (1 year, 9 months)</td>
<td></td>
</tr>
<tr>
<td>Itoh et al (1992)</td>
<td>1</td>
<td>53/M</td>
<td>Parotid gland</td>
<td>Surgery</td>
<td>NR</td>
<td></td>
</tr>
<tr>
<td>Grenko et al (1993)</td>
<td>1</td>
<td>66/F</td>
<td>Parotid gland</td>
<td>Surgery</td>
<td>DWD (13 months after surgery)</td>
<td></td>
</tr>
<tr>
<td>Donath et al (1997)</td>
<td>1</td>
<td>82/M</td>
<td>Parotid gland</td>
<td>NR</td>
<td>NR</td>
<td></td>
</tr>
<tr>
<td>Snyder et al (2000)</td>
<td>1</td>
<td>71/M</td>
<td>Parotid gland</td>
<td>NR</td>
<td>NR</td>
<td></td>
</tr>
<tr>
<td>Tse et al (2004)</td>
<td>1</td>
<td>75/M</td>
<td>Parotid gland</td>
<td>NR</td>
<td>NR</td>
<td></td>
</tr>
<tr>
<td>Tuluc et al (2007)</td>
<td>1</td>
<td>32/F</td>
<td>Nasal Cavity</td>
<td>Surgery</td>
<td>NED (1 year)</td>
<td></td>
</tr>
<tr>
<td>Kadivar et al (2007)</td>
<td>1</td>
<td>75/M</td>
<td>Parotid gland</td>
<td>Surgery</td>
<td>NR</td>
<td></td>
</tr>
<tr>
<td>Fang et al (2009)</td>
<td>1</td>
<td>43/M</td>
<td>Parotid gland</td>
<td>Surgery</td>
<td>NED (1 year)</td>
<td></td>
</tr>
<tr>
<td>Kusafuka et al (2010)</td>
<td>1</td>
<td>40/M</td>
<td>Soft Palate</td>
<td>Surgery</td>
<td>NED (2 years)</td>
<td></td>
</tr>
<tr>
<td>Tanaka et al (present case)</td>
<td>1</td>
<td>37/F</td>
<td>Mental Region</td>
<td>Surgery</td>
<td>NED (5 years and 5 months)</td>
<td></td>
</tr>
</tbody>
</table>

DWD: died with disease, F: female, M: male, NED: no evidence of disease, NR: not recorded

Discussion

Giant cell tumor of soft tissue (GCT-ST), which is extremely rare compared to bony counterparts of GCT, has traditionally been considered a single entity such as tenosynovial GCT, malignant GCT, and other giant cell rich sarcoma, which represented both benign and malignant varieties. Recently GCT-ST is interpreted as the soft tissue analogue of giant cell tumor of bone because of their histological and immunohistochemical similarity. As reported in the recent review, most cases of the GCT-ST are arose from extremities involving either superficial or deep soft tissue. From our review of all the English-language literature, seventeen cases, including our case, were identified as GCT-ST arose in the oral and maxillofacial region. About 82 % (14/17) have occurred in the parotid gland, most of which were associated with a carcinomatous component. To the best our knowledge, no case has been reported, which originate from oral soft tissue region like our case.

To ensure the soft tissue origin of GCT at mental region, we examined the present case by its clinical course, roentgenographic and histological findings. From the roentgenographic perspective, the hyperdense contrast enhancing mass lesion with sharp, dome-shaped, well-defined border was identified at subcutaneous mental region under MR imaging, which was superior to computed tomography (CT) in demonstrating areas of tumor extension into the peripheral soft tissue. Partial bone sclerosis with irregular margin of the mental cortical bone, probably due to a periosteal reaction, was observed by lateral cephalogram. Intraosseous destructive lesion was never identified on all imaging sequences. These findings suggested that the tumor mass occurred at peripheral soft tissue of mandibular bone. Further histopathological findings of excised specimen also excluded the possibility of the soft tissue extension of primary giant cell tumor of bone. Microscopically the lesion was mainly located in the soft tissue of the mental region and partial lytic destruction of adjacent cortical bone was identified, leading to the diagnosis of giant cell tumor of soft tissue at mental region. As for the clinical symptom, the skin surface at the expansive mental region showed redness without ulceration in contrast to the case in GCT of the skin. In addition, the patient had no sustained severe trauma before the appearance of a detectable tumor, denying the reactive lesion such as giant cell reparative granuloma.

Regarding the treatment and clinical prognosis, the majority of GCT-ST, including our case, show histologically benign features as mostly observed in the case of GCT of bone and GCT-ST has a lower recurrence rate than bony counterparts, suggesting a much better accessibility of the soft tissues for surgical excision in general as previously reported by others. Nevertheless, clinical behaviors of GCT-ST are not always favorable because of the tumor infiltrative growth characteristics like our case. For this reason, the wide resection is basically recommended for GCT. However, the proximity of critical structures or the cosmetic disturbance and postoperative motor-sensory dysfunction may occasionally prevent complete excision in the case occurred in oral and maxillofacial region. In our case, complete resection of tumor combined with marginal resection of adjacent bone and soft tissue was capable and resulted in less cosmetic or functional disturbance. Although there is no obvious local recurrence or lung metastasis after surgery in the present case, long-term follow up with careful radiographic workup is required because of the clinically aggressive features of GCT.

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