Localized Tendon Sheath Giant Cell Tumor of Temporomandibular Joint Presenting as a Parotid Gland Tumour: a Rare Case and Review of the Literature

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Abstract

Localized tendon sheath giant cell tumour (TSGCT-L), also known as Tenosynovial giant cell tumour (TGCT) is a rare benign tumour occurring in the hand, knee, ankle, shoulder, fingers and rarely affect the temporomandibular joint.

Clinically TSGCT-L may occur at any age, but usually diagnosed in patients between 30 years and 50 years of age and is more common in women.

We conducted a literature review for 8 cases reports related to TSGCT-L of temporomandibular joint (TMJ) and described one additional case report treated in our hospital. LSGCT-L of the temporomandibular joint is a rare entity, the treatment of choice was surgery excision, but careful preoperative planning with Computed tomography (CT scan) and Magnetic resonance image (MRI) were recommended to obtain a successful outcome with as little morbidity as possible.

Tenosynovial giant cell tumour of temporomandibular joint may be presented clinically and radiologically as a primary parotid gland lesion, with the cytology diagnosis using fine needle aspiration biopsy the suspicion of the parotid gland tumour was excluded due to a undiagnostic result of a tumour.

Keywords: Localized Tendon Sheath Giant Cell Tumour; Temporomandibular Joint; Tenosynovial Giant Cell Tumour; Parotid Gland Tumour

Introduction

Giant cell tumours of tendon sheath also known as Tenosynovial giant cell tumours (TGCT) are rare benign tumour arising in the temporomandibular joint and most common in soft tissue neoplasm of hand, ankle, shoulder and fingers.

Tenosynovial giant cell tumour (TGCT) presents in localized and diffuse forms [1,2].

Localized TGCT is more frequent than diffuse TGCT, representing the most common subset of giant cell tumour. Clinically the localized TSGCT may occur at any age but usually diagnosed in patients between 30 and 50 years of age and is more common in women. It may arise in any synovial lined structure but most commonly develops in the synovial sheath of the flexor or tendons of the hands and fingers [2-6]. However a diffuse Tenosynovial giant cell tumour has a tendency to occur in younger individuals, it’s present as a less well-defined

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soft tissue mass, shows locally aggressive growth and has a high recurrence rate [7]. Although the localized Tenosynovial giant cell tumour (TSGCT-L) contains the same cell population as diffuse Tenosynovial giant cell tumour (TSGCT-D) unlike the diffuse form; usually arose in extra-articular location and grows as a well-defined nodule. The earliest description of the disorder, involving a tendon sheath, was attributed to Chassaignar in 1852.

In the medical literature search, it was very rare for these tumours to be encountered in the parotid gland and especially challenging can be considered when involved the temporomandibular joint. However, in this paper, a detailed case report and literature review of a localized form of tendon sheath giant cell tumour and differential diagnosis of this lesion were discussed.

Case Report

A 54-year-old Chinese female was presented in our hospital in March 2017 with a mass in the left mandibular ramus and condyle, that had been increasing in size slowly about 6 months. Her medical history revealed an antecedent trauma on her face, as well as any pain, hearing loss, but only complaining of a tender sensation in the left parotid gland region. Her maximum interincisal opening was restricted to 25 mm and she had a mandibular deviation to the left side during mouth opening. Computed tomography (CT scan) showed a 2.3 cm x 1.5 cm x 1.5 cm in size with homogeneous solid mass, multilobulated in the superficial parotid gland with extension into the deep lobe, mandibular ramus and condyle of the temporomandibular joint (Figure 1). The clinical diagnosis revealed to be a benign tumour of temporomandibular joint or a pleomorphic adenoma of the parotid gland. In addition, a fine needle aspiration (FNA) biopsy was performed and the result was undiagnostic. Radiographically imaging was investigated and show no obvious abnormalities in the bone of mandibular ramus and condyle of TMJ surrounding by a tumour. Submandibular space and bilateral para pharyngeal structure were clear with no abnormal cervical lymph nodes. Imaging diagnosis result was a left temporomandibular joint tumour. Based on these findings, the patient underwent surgery under general anaesthesia, the lesion was approached via the parotidectomy incision. After elevation of a skin flap, the main branches of facial nerve were isolated, then the parotid gland and encapsulated mass surrounding the mandibular ramus and condyle were identified. This lesion was closed to the facial nerve, the distal branches of the facial nerve (buccal, zygomatic and frontal branches) emitting from a tumour were isolated and marked with silk stitches (blue colour see figure 2) for preserving the nerve damage during the resection of a tumour. This tumour did not affect the temporomandibular joint, the coronoid process and no evidence of involvement the zygomatic arch. An intraoperative frozen section was performed with the resection specimen consisted of soft tissue fragment measuring 3.8 cm x 3 cm x 3 cm in aggregate. The cut surface showed the yellow-white colour of the lesion, lobulated and encapsulated with focal haemorrhage (Figure 3). Pathological diagnosis result was localized of tendon sheath giant cell tumour of left mandibular ramus and condyle) (TSGCT-L). All the mass was removed as completely as possible, preserving the temporomandibular structures and facial nerves branches. The patient mouth opening and complaining the tender sensation of the left parotid region were resolved with satisfaction during the one-year follow-up, no recurrence was observed.

Figure 1: Computed tomography (CT scan) axial view showed a 2.3 cm x 1.5 cm x 1.5 cm in size with homogeneous solid mass, multilobulated in the superficial parotid gland with extension into the deep lobe, mandibular ramus and condyle of the temporomandibular joint
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Microscopically, the lesion was showed short-shaped cells scattered, calcification exhibited round to oval nuclei, accompanied by hematoxylin deposition. Numerous multinucleated giant cell tumour dispersed evenly among monocyte-macrophages and spindle cells. The spindle cells represent the neoplastic cells in this tumour, and that the monocyte-macrophages were reactivated giving rise to giant cells (Figure 4).

**Figure 4:** Microscopically the lesion showed short-shaped cells scattered, calcification exhibited round to oval nuclei, accompanied by hematoxylin deposition.

Immunohistochemical staining was used. Most of the multi nuclear and mononuclear exhibited intense cytoplasmic positivity for vimentin, CD68, LCA (leukocyte common antigen) and negative staining for smooth muscle actin (SMA), P63, Cytokeratins (CK, CK7) and CD57. Cytokeratin is an epithelial marker; SMA is used to identify normal and neoplastic smooth muscle cells, myofibroblasts and myoepithelial cells and P63(-) protein is often used to identify myoepithelial and squamous cell neoplasms (Figure 5).

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Figure 5: Immunohistochemical staining was used. Most of the multinuclear and mononuclear exhibited intense cytoplasmic positivity for vimentin, CD68, LCA (leukocyte common antigen) and negative staining for smooth muscle actin (SMA), P63, Cytokeratins (CK, CK7) and CD57.

Based on the histopathological examination and immunohistochemical examination, the final diagnosis was localized tendon sheath giant cell tumour (TSGCT-L) of left (mandibular ramus and condyle of temporomandibular joint).

The management of localized tendon sheath giant cell tumour included surgical resection, radiotherapy, chemotherapy or combination. Here the treatment choice for TSGCT-L of temporomandibular joint was surgical removal of a tumour.

Discussion

In the literature 8 cases of localized tendon sheath giant cell tumours of TMJ have been reported in English literature and were summarized in table, 2 males and 6 females ranging from 21 to 78 years old with a mean age of 50.13 years.

<table>
<thead>
<tr>
<th>Authors (dates)</th>
<th>Ages/Sex (M/F)</th>
<th>Diagnosis</th>
<th>Treatment</th>
<th>Follow-up</th>
<th>Local Recurrence</th>
</tr>
</thead>
<tbody>
<tr>
<td>Raibley (1977) [13]</td>
<td>62yrs/F</td>
<td>TSGCT-L</td>
<td>Surgery</td>
<td>5 months</td>
<td>No</td>
</tr>
<tr>
<td>Malek and Drommer (1978) [14]</td>
<td>55yrs/F</td>
<td>TSGCT-L</td>
<td>Surgery</td>
<td>9 months</td>
<td>No</td>
</tr>
<tr>
<td>Youssef., et al. (1996) [15]</td>
<td>41yrs/F</td>
<td>TSGCT-L</td>
<td>Surgery</td>
<td>14 months</td>
<td>No</td>
</tr>
<tr>
<td>Allias –Montmayeur., et al. (1997) [16]</td>
<td>39yrs/F</td>
<td>TSGCT-L</td>
<td>Surgery</td>
<td>7 months</td>
<td>No</td>
</tr>
<tr>
<td>Cascone., et al. (2005) [17]</td>
<td>38yrs/F</td>
<td>TSGCT-L</td>
<td>Surgery</td>
<td>1 year</td>
<td>No</td>
</tr>
<tr>
<td>Izzo., et al. (2005) [18]</td>
<td>67 yrs/M</td>
<td>TSGCT-L</td>
<td>Surgery</td>
<td>1 months</td>
<td>No</td>
</tr>
<tr>
<td>Gascone., et al. (2008) [19]</td>
<td>78yrs/M</td>
<td>TSGCT-L</td>
<td>Surgery</td>
<td>2 year</td>
<td>No</td>
</tr>
</tbody>
</table>

Table: Summary of the 8 cases of TSGCT-L of the temporomandibular joint (TMJ).

Surgical excision of a tumour was a successful treatment of the all patients with localized tendon sheath giant cell tumour. No local recurrence of these cases during the following – up with an average of 10.89 months. In the medical search, the localized form has been
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regarded as giant cell tumour of tendon sheath (GCTTS) as they characteristically affect the synovial tendon sheath of hands, and feet [8,9]. TSGCT-L is the most common soft tissue neoplasm of the hand, clinically the patients presented with a mobile firm nodular mass that is usually less than 2.0 cm in diameter. On imaging studies, TSGCT-L appears as soft tissue mass that was associated with bone erosions in approximately 18% - 20% of cases. The erosions are well defined and have sclerotic margins caused by the direct pressure of a tumour on the underlying bone. Their appearance on MRI was variable depending on the amount of hemosiderin deposition. The lesion was intermediate in signal intensity on T1 weighted images, intermediate to low signal on T2-weighted images and show moderate to intense enhancement on gadolinium- enhanced images. Grossly, TSGCT-L was lobulated, rubbery and well-circumscribed cross-sections reveal a mass that was grey-tan with brown and yellow areas. In this case presented the resection specimen showed in the cut surface a yellow –white colour of a tumour, lobulated and encapsulated tissue with focal haemorrhage. Microscopically, TSGCT-L was identical to the individual lobules seen in TSGCT-Diffuse [3,4]. Although there may be less hemosiderin and more extracellular collagen. Surgical excision was the standard form of treatment and the local recurrence was approximately 10% - 20%. The author case reported the local recurrence did not observe, during the one-year follow-up, the patient obtained satisfaction of treatment. The distinction between TSGCT-L and TSGCT-D can be difficult to make on a small biopsy and the histologic features must be correlated with the clinical or radiographic findings.

The diagnosis can also be difficult in cases where the needle biopsy sample areas of sclerosis or cellular areas devoid of multinucleated giant cells. Occasionally, small clusters of osteoclast–type giant cells can be seen in inflamed synovium. These cells were few and sub synovial in location and should not be confused with TSGCT-D. The localized variant known as giant cell tumour of tendon sheath and localized nodular synovitis forms a discrete nodule and primarily affects the Tenosynovium of the hand. However, the diffuse form known as pigmented villonodular synovitis involved the synovium in a more diffuse fashion and affected large weight bearing joints such as the knee. Supporting the concept that these lesions are neoplastic, the studies F1 expression in the neoplastic cells. Although CSF1 was only expressed in the neoplastic mononuclear cells. It's receptor CSF1B was expressed in all cells types seen in the TSGCT. Both types of TSGCT were microscopically identical and over express colony stimulating factor (CSF) [10].

The author case reported, the fine needle aspiration (FNA) biopsy of this tumour was undiagnostic, this was excluded the parotid gland tumour because the cytology biopsy of fine needle aspiration has been a very powerful tool in the diagnosis of primary salivary gland lesions. Radiologically as a primary parotid gland lesion, using a fine needle aspiration biopsy, the suspicion of parotid gland tumour was excluded. For differential diagnosis, common benign tumours occurring to the TMJ included osteochondroma, giant cell granuloma, osteoma, myxoma, synovial chondromatous and hemangioma of the capitulum [11].

Based on the histopathological examination and immunohistochemical examination, the final diagnosis was localized tendon sheath giant cell tumour of the left temporomandibular joint. The treatment was surgical resection of a tumour.

However, tumors that involved the TMJ are very rare and may originate in the joint or invade the TMJ area from outside. Malignant tumours such as fibrosarcoma, chondrosarcoma, synovial sarcoma and Langerhans cell histiocytosis can also be occurring in the TMJ[12].

Conclusion

In conclusion, tenosynovial giant cell tumour of the temporomandibular joint may be present clinically and radiologically as a primary parotid gland tumour with the cytology using fine needle aspiration biopsy the result shows a undiagnostic tumour that excluded the parotid gland tumour.

Bibliography


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