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Case report

Giant cell tumour of a temporomandibular joint presenting as a parotid mass: Case report and analysis of the 19 cases in the literature

Yun-Chen Huang [a], Jeng-Wen Chen [a], Yen-Lin Chen [b], Pei-Jen Lou [c, *]

[a] Department of Otolaryngology, Head and Neck Surgery, Cardinal Tien Hospital, School of Medicine, Fu-Jen Catholic University, Taipei, Taiwan
[b] Department of Pathology, Cardinal Tien Hospital, School of Medicine, Fu-Jen Catholic University, Taipei, Taiwan
[c] Department of Otolaryngology, Head and Neck Surgery, National Taiwan University Hospital, School of Medicine, National Taiwan University, Taipei, Taiwan

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ABSTRACT

Giant cell tumour (GCT) in the parotid region is a rare lesion whose origin can be divided into three compartments: giant cell tumour of the salivary gland (GCT-SG), giant cell tumour of the bone (GCT-bone) and giant cell tumour of the soft tissue (GCT-ST). A low risk of malignancy has been observed, in which all of them were GCT-SG.

We present a case of GCT in the parotid region and review the features of GCT in origin, immunohistochemical characteristics, treatment of choice and disease outcome.

1. Introduction

A parotid mass is a common presenting complaint for head and neck surgeons. The most common benign parotid tumour is the pleomorphic adenoma (Pinkston and Cole, 1999). Some rare extraparotid lesions, arising from the temporomandibular joint or mandible, may also present as a parotid mass and should be considered in the differential diagnosis. Histologically characterized as round to oval-shaped mononuclear cells (MNC) intimately admixed with uniformly scattered multinucleated osteoclast-like giant cells (MOGC), GCT is generally considered a benign tumour originating from the undifferentiated mesenchymal cells of the bone marrow; it is most commonly found in the extremities (Mendenhall et al., 2006; Sethi et al., 2006). Three types of GCT are GCT of the bone, GCT of the tendon sheath and GCT of the soft tissue. Notably, GCT located in the head and neck region is uncommon (Roy et al., 2013). Around the parotid region, only eighteen cases have been reported in English-language literature (Eusebi et al., 1984; Balogh et al., 1985; Batsakis et al., 1988; Ellis et al., 1991; Itoh et al., 1992; Grenko et al., 1993; Donath et al., 1997; Tse et al., 2004; Torabinezad et al., 2006; Kadivar et al., 2007; Fang et al., 2009; Wu et al., 2012; Yang et al., 2012). According to the anatomic location, GCT in the parotid region can be divided into intraparotid and extraparotid lesions. GCT arises mainly from the parotid gland (i.e., giant cell tumour of the salivary gland, GCT-SG) and is regarded as the intraparotid lesion, but may also arise in the mandible (i.e., giant cell tumour of the bone, GCT-bone), or the soft tissue in the parotid region (i.e., giant cell tumour of the soft tissue, GCT-ST) regarded as extraparotid lesions (Tse et al., 2004; Fang et al., 2009). Ten of the eighteen reported cases were malignant, and all originated from GCT-SG (Eusebi et al., 1984; Balogh et al., 1985; Batsakis et al., 1988; Grenko et al., 1993; Donath et al., 1997; Tse et al., 2004; Torabinezad et al., 2006; Kadivar et al., 2007; Fang et al., 2009; Yang et al., 2012). This finding highlights the importance of the origin of GCT in the parotid region. Given the rarity of the above cases, no consistent treatment guideline has been proposed. Adjuvant radiotherapy was performed in two cases, possibly owing to the radiosensitive nature of the giant cell tumour (Balogh et al., 1985; Kanamori and Ohmori, 2005; Wu et al., 2012). Based on the case study reported in this study, the 19th case of GCT in the parotid region is demonstrated and related literature is reviewed as well.

2. Case report

A 41-year-old man presented to our clinic with a non-progressive, painless and indurate mass in the right parotid region for five months. The lesion was traced back to seven months

* Corresponding author. Department of Otolaryngology, Head and Neck Surgery, National Taiwan University Hospital and National Taiwan University School of Medicine, 7, Chung-Shan South Road, Taipei, Taiwan. Tel.: +886 2 23123456x65224; fax: +886 2 23410905.
E-mail addresses: pjlou@ntu.edu.tw, apjlou@gmail.com (P.-J. Lou).

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before admission when the right parotid mass was noted. He first received partial excision of the right parotid mass at another hospital. The pathological diagnosis was GCT-ST. Postoperatively, induration was still found in the right parotid area. The patient came to our hospital for a second opinion, when physical examination revealed a 2 × 2 cm painless indurate mass in the right parotid area with a previous surgical scar. Examination of the head and neck, including nasopharynx, oral cavity, hypopharynx, and larynx were unremarkable. Routine blood chemistry tests were normal. Magnetic resonance imaging revealed a 2 cm mass with T1W isosignal and T2W hyposignal adjoining to the mandible bone in the area corresponding to the parotid gland on the right side (Fig. 1). Heterogeneous enhancement of the mass was noted.

A skin incision was made from anterior to the right ear to the mandibular margin. The tumour was extraparotid and was located beneath the zygomatic and temporal branches of facial nerves, firmly adhering to the temporomandibular joint capsule (Fig. 2A). The tumour was then resected completely along with a portion of the temporomandibular joint capsule. Notably, the tumour did not involve the mandible. The zygomatic and temporal branches of facial nerve were well preserved (Fig. 2C). Postoperatively, facial palsy was mild and gradually improved.

Gross examination revealed a brown and elastic mass, measuring up to 2.5 × 1.2 × 0.9 cm in size (Fig 3A). The microscopic examination in Fig. 3B indicated a giant cell-rich lesion characterized by a multinodular architecture, with cellular nodules separated by fibrous tissue septa. The nodules consisted of a mixture of round to oval cells that were MNC and MOGC, both of which had similar nuclei and were immersed in richly vascularized stroma. Immunohistochemically, CD68 highlighted the MOGC and a minor portion of MNC (Fig. 3C). S-100 and cytokeratin were both negative (data not shown). The margin was free of tumour involvement.

The diagnosis was GCT-ST (the temporomandibular joint capsule) in the right parotid region. The patient had no facial palsy and no parotid mass at 14-months follow up examination.

3. Discussion

Neoplasms in the parotid region arise mainly from the parotid gland and are usually benign. The most common tumour is pleomorphic adenoma, accounting for approximately 53.3% of all parotid gland neoplasm. It is followed by Warthin tumour, which accounts for approximately 28.3% of all parotid gland neoplasm. Only 14.6% of the parotid gland neoplasms are malignant (Pinkston and Cole, 1999). Some rare extraparotid lesions, originating from the surrounding tissue of the parotid gland (e.g., the temporomandibular joint or mandible) also present as a parotid mass and should be considered in the differential diagnosis because the incidence of malignancy and treatment might differ from other lesions. Eusebi et al. (1984) first reported three cases of GCT in the parotid region with an unclear origin. Histologically characterized as a round to oval-shaped MNC intimately admixed with uniformly scattered MOGC, GCT is generally considered a benign tumour originating from the undifferentiated mesenchymal cells of the bone marrow; this tumour is most commonly found in the extremities (Mendenhall et al., 2006; Sethi et al., 2006). GCT outside the bone is histologically similar to GCT of the bone, and possibly occurring in the soft tissue (Trabelsi et al., 2009) and in visceral organs such as the pancreas (Silverman et al., 1990), thyroid (Cibull and Gray, 1978), and liver (Matsumoto et al., 2012). Therefore, we can infer that GCT can arise from the bone or soft tissue adjacent to the parotid gland. Including the current case, only nineteen cases of GCT located in the parotid region have been reported in the English-language literature. In addition to reporting this rare case, we also demonstrated the surgical intervention rarely found in the literature (Itoh et al., 1992).

As shown in Table 1, most patients (16/19) reviewed in the literature were males, with their ages ranging from 30 to 92 years. Ten cases were found to be malignant. Of the malignant components within GCT, six cases were salivary duct carcinoma (Balogh et al., 1985; Batsakis et al., 1988; Grenko et al., 1993; Tse et al., 2004; Kadivar et al., 2007; Fang et al., 2009); two cases were carcinoma ex pleomorphic adenoma (Eusebi et al., 1984; Donath et al., 1997); and two cases revealed carcinoma without definite pathological diagnosis (Torabinezad et al., 2006; Yang et al., 2012). All malignant components within GCT in the parotid region were carcinoma. Cervical lymph node metastasis was present in one case (Kadivar et al., 2007). Additionally, lung metastasis was reported in two cases, and both of them died with disease (Balogh et al., 1985; Grenko et al., 1993). Most cases received surgical intervention alone with only two cases receiving adjuvant radiotherapy (Balogh et al., 1985; Wu et al., 2012). No recurrence case was reported.

Although GCT of the bone or soft tissue can harbour a malignant tumour, the malignancy is invariably sarcoma rather than carcinoma (Bertoni et al., 2003; Tse et al., 2004). However, all malignant components within GCT in the parotid region were carcinoma. Additionally, in certain malignant cases with salivary duct carcinoma, previous studies demonstrated that the carcinomatous component has genotyping analysis similar to that of mononuclear cells (Balogh et al., 1985; Batsakis et al., 1988; Grenko et al., 1993; Tse et al., 2004; Kadivar et al., 2007; Fang et al., 2009). This finding suggests a possible origin from the parotid gland. In two

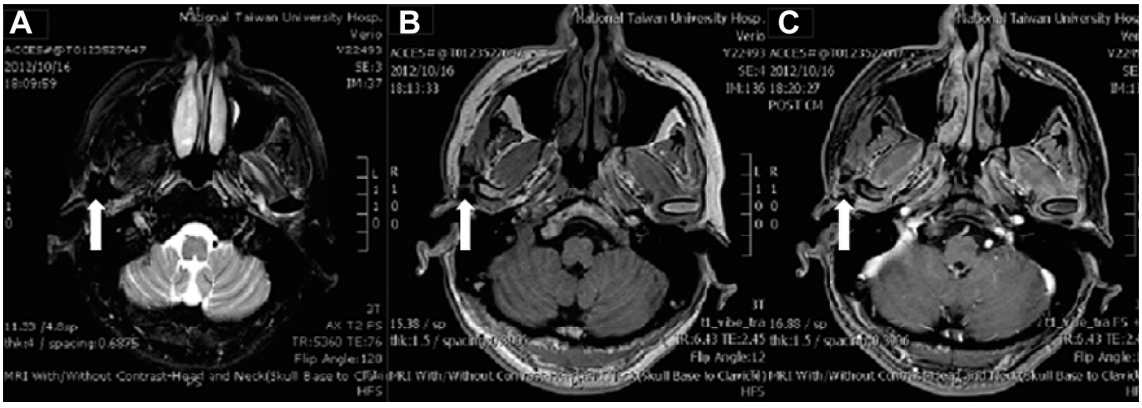

Fig. 1. Magnetic resonance imaging findings. The mass, measuring 2 cm in size, adjoining to the mandible condyle was hyposignal in T2W (A) and isosignal in T1W (B). Heterogeneous enhancement of the mass in T1W with contrast (C).