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Review Article

Giant-cell tumors of soft tissue in the head and neck: A review article

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ABSTRACT

Giant-cell tumor of soft tissue (GCT-ST) is a rare neoplasm that was first described in 1972. Due to its histological and immune-histochemical similarities with GCT of bones, GCT-ST is thought to be its counterpart. This review article aims to recognize the characteristics of this rare tumor along with a favorable way to diagnose and treat. We reviewed 12 cases of GCT-ST in the head and neck. This tumor involves both sexes at any age. Histopathology in many cases revealed the presence of mononuclear cells and spindle cells. Immunohistochemistry was positive in many cases for CD68. Complete excision was the most accepted management technique; however, few cases reported recurrence and metastasis. Post-operative radiology is suggested to prevent local recurrence and in case of incomplete excision of the tumor.

Keywords: Giant-cell, head and neck, tumor, review

Introduction

Giant-cell tumor of soft tissue (GCT-ST) is a rare neoplasm that was first described in 1972.[1,2] Due to its histological and immunohistochemical similarities with GCT of bones, GCT-ST is thought to be its counterpart.[3] The World Health Organization classifies the tumor as a GCT-ST with low malignant potential (GCT-LMP) and malignant GCT-ST.[4-6] GCT-ST is characterized by the presence of spindles or polygonal mononuclear cells and multinucleated osteoclast-like giant cells.[2] Atypia and necrosis are present in the malignant form of GCT-ST. GCT-LMP is most commonly found in superficial soft tissue of upper and lower extremities, especially on hands, arms, and feet.[1,4,7-13] The head and neck region is rarely affected.[3] In most patients, the chief complaint is a painless well-circumscribed mass covered by normal skin or by a fleshy red-brown surface when superficially located.[2,13,14] The duration of the mass varies between 2 and 12 months.[2] There is no predominant age group affected by the tumor specifically, as it occurs in a broad age range from 5 to 84 years with no sex predilection.[1,4,7-12,15] No exact etiological factors have been identified.[6] Differential diagnosis should include benign lesions (such as GCT of tendon sheaths, cellular dermatofibroma with osteoclast-like giant cells, ossifying dermatofibroma with osteoclast-like giant cells, reparative giant-cell granuloma, nodular fasciitis, and brown tumor of hyperparathyroidism extending to soft tissues) and malignant lesions (leiomyosarcoma with osteoclast-like giant cells, epithelioid sarcoma with giant cells, extraskeletal osteosarcoma, atypical fibroxanthoma with osteoclast-like giant cells, and plexiform fibrohistiocytic tumor).[2,16-18] Cytopathology, histopathology, and immunostains for smooth muscle actin and CD68 are important for confirming the diagnosis and ruling out neoplastic and non-neoplastic differential diagnoses.[3]

Complete excision with a free surgical margin is thought to be the best treatment approach.[3,9,15,19] There have been a few reported cases of local recurrence and metastasis with positive surgical margins.[20] As GCT-ST is radiosensitive,[1,21] radiotherapy should be considered for cases with incomplete surgical margin excision due to its proximity to critical structures.[15,16]

This tumor has biologically benign behavior,[17,18] and thus, removal of the tumor with clear surgical margins guarantees no recurrence.[22] However, strict follow-up is recommended, as local recurrence and metastasis cannot be predicted.[13,14] This review article aimed to recognize the characteristics of this rare tumor along with favorable ways to diagnose and treat it.

Results and Discussion

Literature reviews of GCT-ST cases were identified between 1981 and 2016 using the search engines PubMed, Ovid, and Medline. The keywords were as follows: GCT-ST and GCT-LMP. We included all cases of GCT-ST in the head and neck region. The language was limited to English, and we excluded GCT of bones, cartilages, other body regions, and glands.

We reviewed 12 cases, as described in Table 1. All cases are arranged chronologically from 1981 to 2016. We searched for study type, year of publication, patient demographics, initial presentation, and the tumor site. We focused on the tumor features by discussing the physical examination findings and radiological test reports if applicable, while all microscopic examinations data were collected. Additional tests included immunohistochemical staining, in addition to management plans and follow-up outcomes.

Most patients, 7 of them, were male (58.3%)[3,9,10,20,23-25] and 5 (41.7%)[13,15,26-28] were female. This tumor involves a wide

Table 1: GCT cases

Study	Type	Year	Patients age in years and gender	Initial presentation	Tumor site	Characteristic features on microscopic examination	Immunohistochemistry positive results	Management and follow-up
Angervall *et al*.[10]	Retrospective	1981	75/M	Painless mass	Left neck	Mononuclear spindle cells and osteoclast-like giant cells Malignant pattern was observed	N/A	Local excision Died after 1 year of metastases
Oliveira *et al*.[9]	Retrospective	2000	9/M	-	Left neck	Mononuclear spindle cells and osteoclast-like giant cells	N/A	Lost to follow-up
Hoang *et al*.[23]	Case series	2002	73/M	Asymptomatic exophytic nodules.	Right forehead	Multinucleated giant cells	CD68 Alpha-1 -antichymotrypsin	Local excision Lost to follow-up
Tuluc *et al*.[26]	Case report	2006	32/F	Frequent sinus infection and nasal obstruction	Intranasal mass	Mononuclear spindle cells and osteoclast-like giant cells Brisk mitotic rate was noted but no atypical forms	CD68 TRAP	Lost to follow-up No recurrence
Pepper *et al*.[15]	Case report	2009	59/F	Painless mass	Left parotid	Mononuclear spindle cells and osteoclast-like giant cells	CD68	Parotidectomy Postoperative radiotherapy
Trabelsi, *et al*.[24]	Case report	2009	28/M	Painless mass	Left posterior neck	Mononuclear spindle cells and osteoclast-like giant cells	CD68	Local excision No recurrence
Calli *et al*.[27]	Case report	2011	53/F	Painless mass	Left neck mass	Osteoclast-like giant cells Mild nuclear atypia Hemorrhagic areas were occasionally seen	CD68 Vimentin Alpha-1 -antitrypsin	Local excision Post-operative radiotherapy No recurrence
Kumar *et al*.[25]	Letter to editor	2013	22/M	Intermittent headache over the left temporal area, associated with progressive diminution of vision in both eyes	Left temporal area	Mononuclear spindle cells and osteoclast-like giant cells	N/A	Subtotal resection
Righi *et al*.[13]	Case report	2014	36/F	Painless cystic mass	Inferior lip	Mononuclear spindle cells and osteoclast-like giant cells Mild to moderate nuclear atypia	CD68 Vimentin Actin	Local excision Recurrence occur on the anterior tibia and excised

(Contd...)

Table 1: *(Continued)*

Study	Type	Year	Patients age in years and gender	Initial presentation	Tumor site	Characteristic features on microscopic examination	Immunohistochemistry positive results	Management and follow-up
Bandyopadhyay *et al*.[3]	Case report	2015	35/M	Painless mass	Right submandibular region	Mononuclear spindle cells and osteoclast-like giant cells	CD68 Smooth muscle actin (SMA)	Local excision No recurrence
Devrim T.[20]	Case report	2015	65/M	Painless mass	Superior lip.	Mononuclear spindle cells and osteoclast-like giant cells	CD68	Local excision
Hafiz, *et al*.[28]	Case report	2016	28/F	Painless mass with otorrhea	Right ear	Mononuclear spindle cells and osteoclast-like giant cells The mononuclear cells exhibited brisk mitotic activity	N/A	Local excision

GCT: Giant-cell tumor

age range from 9 to 75 years, but more commonly affects middle-aged individuals. For the site, 7 (58.3%) cases involved multiple sites in the head: Forehead, temporal area, parotid, ear, nose, superior, and inferior lip.[13,15,20,23,25-28] In the neck region, however, 5 (41.7%) cases were located in different neck triangles.[3,9,10,24,27] The main complaint was a painless growing mass in 9 (75%) cases.[3,9,10,13,15,20,23,24,27] However, 3 (25%) cases reported different complaints based on the site of the tumor including sinus infection, otorrhea, loss of vision, and headache.[25,26,28] Most cases described the mass as slow-growing, well-circumscribed, and covered by normal skin, while only 1 case had pus and discharge, and another case had an exophytic mass.[23,28] The mass sizes ranged between 15 mm and 5 cm. The duration of symptoms was between 1 month and 1 year. For radiological examination, computed tomography scan showed no relation to the bone. Microscopic examination revealed most of the tumors were composed of osteoclast-like multinucleated giant cells, mononuclear cells, and spindle cells in 6 (50%) cases. In almost a third of the cases, 4 subjects (33.3%) exhibited a mild to moderate degree of nuclear atypia and nuclear pleomorphism,[10,13,26,28] which are indicators of malignancy. The osteoclast-like giant cells showed strongly positive CD68 in 9 (75%) cases[3,13,15,20,23-27] and were negative for CD34 in 3 (25%) cases.[13,15,20] Surgical excision, which is the most accepted management technique, was done for all cases except for one that was managed by subtotal resection.[25] All cases were followed up except for 2 cases.[9,23] Metastasis to the lung was reported in 1 case, and the patient died 1 year later.[10] Recurrence rate was very low; specifically, 1 case had recurrence after 6 months of resection in the tibia.[13] Post-operative radiotherapy was given for 2 (16.7%) cases, one of which reported that the period of its course was over 5 weeks with no evidence of recurrence and metastasis.[15,27] There are no definitive studies supporting post-operative radiotherapy; however, it is suggested because of the possibility of recurrence, especially for cases with incomplete excision.

Conclusion

We reviewed 12 cases of GCT-ST in the head and neck. This tumor involves both sexes at any age. Histopathology in many cases revealed the presence of mononuclear cells and spindle cells. Immunohistochemistry was also positive in many cases for CD68. Complete excision was the most accepted management. However, few cases reported recurrence and metastasis. Post-operative radiology is suggested to prevent local recurrence and in case of incomplete excision of the tumor.

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