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Case Report

Giant cell tumor of the tendon sheath arising from a membrane surrounding the posterior arch of C1: a case report

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Abstract

BACKGROUND CONTEXT: Giant cell tumor of the tendon sheath (GCTTS) is a common, benign lesion of the synovial membrane that occurs more often in large joints than in digits. Giant cell tumor of the tendon sheath rarely arises in close proximity to the axial skeleton.

PURPOSE: The purpose of the study was to report a rare case of GCTTS arising from the membrane surrounding the posterior arch of the atlas (C1).

STUDY DESIGN/SETTING: This is a case report.

METHODS: The methods involve clinical findings and review of current literature.

RESULTS: In this report, we describe a rare case of GCTTS arising from the membrane surrounding the posterior arch of C1, with no apparent continuity with the facet joint. Here we show the radiographic features, with particular emphasis on positron emission tomography-computerized tomography scans, which have not been previously reported.

CONCLUSIONS: We experienced an extremely rare case of GCTTS arising from the membrane surrounding the posterior arch of the C1 vertebra. In spite of the rarity of this disease, GCTTS should be considered in the differential diagnosis of the axial skeletal lesion. Awareness of GCTTS is important because its radiographic features may simulate other neoplastic lesions in the spine.

Introduction

Giant cell tumor of the tendon sheath (GCTTS) belongs to a group of common benign lesions arising from the synovium of tendon sheaths, bursas, and joints. Based on their locations and encapsulation, they are classified as localized or diffuse. Diffuse GCTTS, also called pigmented villonodular synovitis (PVNS), typically affects the synovium of large joints, such as the knee, hip, ankle, and elbow. Localized GCTTS usually affects the tendons of the hands and feet, although it can be apparent in the large joints. The occurrence of GCTTS in the axial skeleton is very rare. Axial involvement in patients predominantly occurs in the lumbar or cervical spine [1]. The spinal form of GCTTS is thought to arise from the synovial membrane of the facet joints [2].

In this report, we describe a rare case of GCTTS arising from the membrane surrounding the posterior arch of the atlas (C1), with no apparent continuity with the facet joint. In the present study, we show the radiographic features, particularly positron emission tomography-computerized tomography (PET-CT) scans, which have not been previously reported for such a tumor.

FDA device/drug status: Not applicable.

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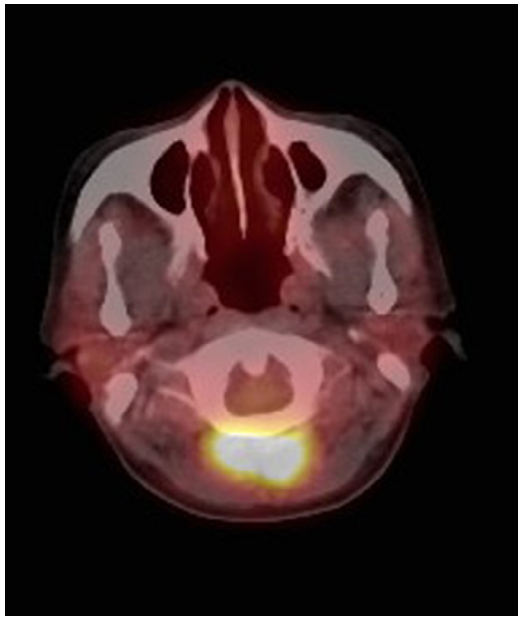

Fig. 1. Positron emission tomography-computerized tomography (PET-CT) scan showed a soft tissue mass (SUVmax: 10.10) behind the posterior arch of C1.

Case report

Presentation and examination

A 63-year-old woman who was previously asymptomatic except for occasional photopsia underwent a PET-CT scan for health screening. A soft tissue mass (standardized uptake value max [SUVmax]: 10.10) was incidentally discovered behind the posterior arch of C1 (Fig. 1); however, no masses were found on palpation. Neurologic testing failed to detect any functional deficits in the upper and lower extremities, and the patient had a full range of motion of the cervical spine without associated pain. Laboratory investigations revealed normal hematologic and biochemical parameters. Radiographs showed only slight spondylosis of the upper cervical spine. Contrast CT showed a 30-mm lobulated mass with slight contrast enhancement without expansion or thinning of the cortex (Fig. 2). There were no soft tissue calcifications. Magnetic resonance imaging (MRI) demonstrated a large, lobulated mass extending from the occipitalis (C0) to the axis (C2) around the posterior arch of C1 to involve the paraspinous muscles. The mass extended into the spinal canal slightly but was not connected to the facet joints. On T1-weighted images, this lesion was isointense with muscles. On T2-weighted images, the mass exhibited heterogeneous signal intensity that showed slightly increased or decreased signal intensity compared with muscles (Fig. 3). This lesion was of low signal intensity on both T1-weighted fat-suppressed images (short TI inversion recovery, STIR) and diffusion-weighted images. These images also demonstrated that the mass was separate from the facet joints. Thallium scintigraphy was performed to exclude malignancy. The mass did not exhibit increased uptake. Based on the clinical and radiological findings, the differential diagnosis for this lesion was GCTTS, desmoid, fibroma, or another benign or malignant lesion.

Surgical findings

Because the imaging studies could not rule out a malignancy, the patient agreed to the surgical excision of the lesion. An open biopsy was performed, and examination of the initial frozen section revealed no malignancy. The lesion was then entirely removed. The tumor was composed of rubbery, soft, yellowish-brown tissue coated by a thin film (Fig. 4). The tumor was firmly attached to the posterior occipitoatlantal membrane. No continuity was observed between the lesion and the facet joints.

Pathologic findings

Microscopic examination of permanent histologic sections showed that the tumor consisted of histiocyte-like mononuclear, foamy, and multinuclear giant cells. Collagenous fibers infiltrated the interstitial space. The histologic findings were compatible with a diagnosis of GCTTS (Fig. 5). Based on imaging and intraoperative features, the lesion was strongly suspected of arising from the posterior occipitoatlantal membrane.

Postoperative period

After surgery, the patient made an uneventful recovery. The occasional photopsia completely disappeared. At the most recent follow-up examination, 3 years after surgery, MRI revealed no evidence of recurrence.

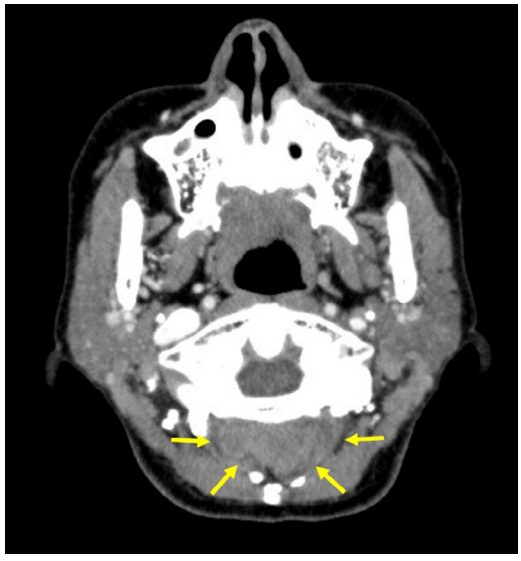

Fig. 2. Contrast computed tomography (CT) showed a 30-mm lobulated mass with slight contrast enhancement without expansion or thinning of the cortex.

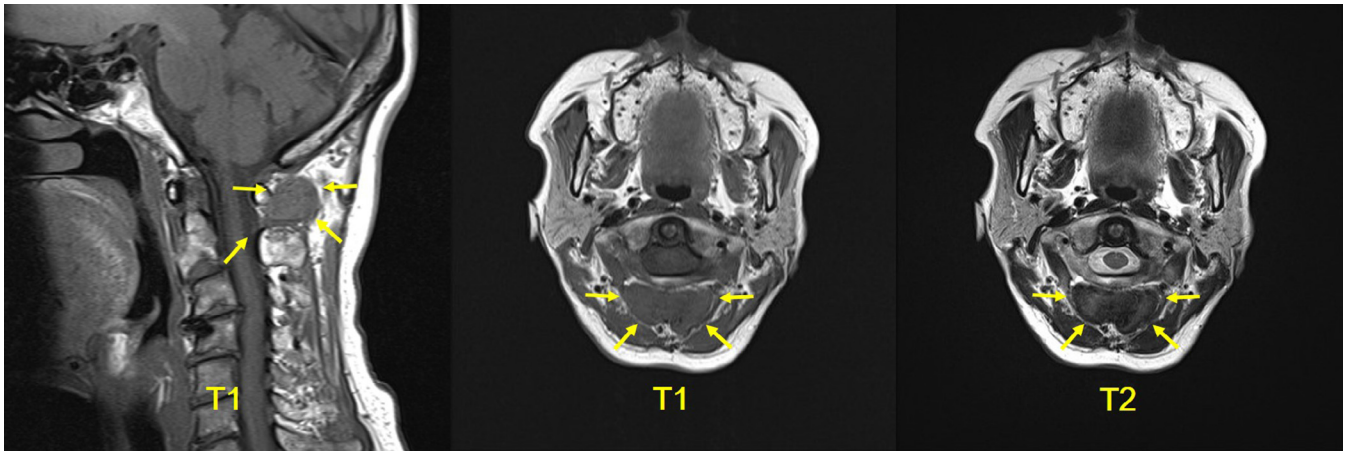

Fig. 3. Magnetic resonance imaging (MRI) demonstrated a large, lobulated mass extending from C0 to C2 around the posterior arch of C1 to involve the paraspinous muscles. The mass extended into the spinal canal but did not continue to the facet joints or the occipital condyles. On T1-weighted images, this lesion was isointense to muscles; on T2-weighted images, the mass exhibited heterogeneous signal intensity with slightly increased or decreased signal intensity compared with muscles.

Discussion

Giant cell tumor of the tendon sheath and PVNS are histologically and chromosomally the same disorders with different anatomical origins and clinical presentations. These diseases were first described by Jaffe et al. [3] as reactive synovitis. The main difference between PVNS and GCTTS is that the former is mainly an intra-articular growth, whereas the latter predominantly grows outside the joints. Giant cell tumor of the tendon sheath most commonly occurs in the hands and occasionally involves the ankles, toes, knees, and hips [4]. Giant cell tumor of the tendon sheath only rarely arises in close proximity to the axial skeleton; with respect to anatomical location, 52% involve the cervical spine, 29% involve the lumbar spine, and 17% involve the thoracic spine [5]. The upper cervical spine is a rare site for the occurrence of GCTTS, and only five cases have been described to date [6–10].

The most commonly reported origin of this tumor is the synovial membrane of the facet joints and bursas. According to Enzinger and Weiss [11], the exact site of the origin of these lesions is often difficult to determine. The previous four cases located in the upper cervical spine originated in the joints [6–8,10], but in only one case did the tumor presumably rise within a small bursa [9]. In our case, the lesion

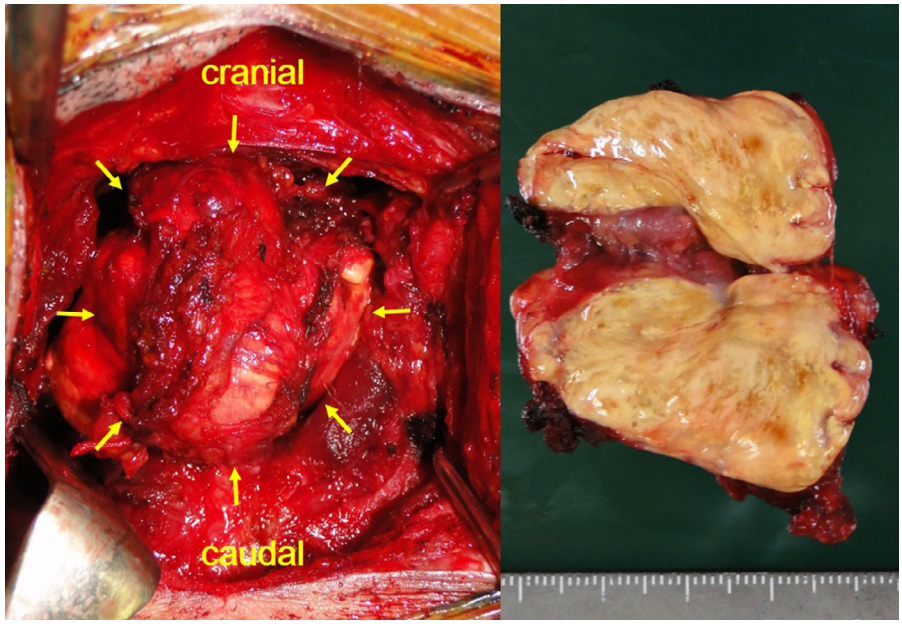

Fig. 4. Resected tumor. The tumor was a soft, rubbery, yellowish-brown tissue and coated by a thin film. It was attached firmly to the posterior occipitoatlantal membrane. No continuity was found between the lesion and the facet joints.

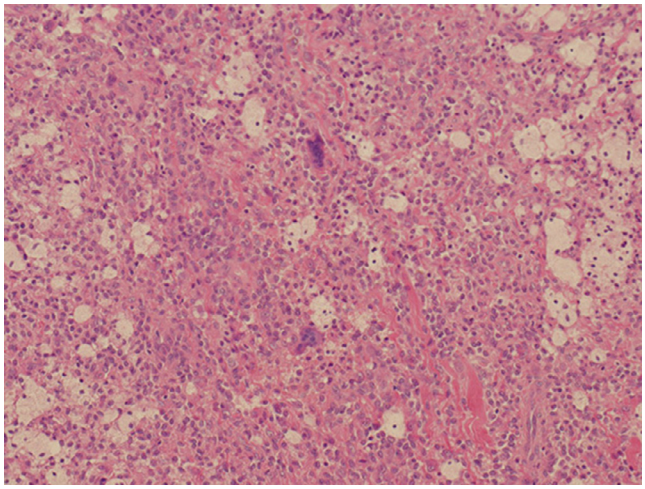

Fig. 5. Pathology. The tumor included histiocyte-like mononuclear cells, foamy cells, small numbers of multinuclear giant cells, and collagenous fibers infiltrating the interstitial space.

most likely arose from the membrane around the posterior arch of C1, with no apparent continuity with the facet joints.

Giant cell tumor of the tendon sheath arises extra-articularly as a slow-growing mass that is well circumscribed and causes little discomfort [12]. Giant cell tumor of the tendon sheath of the spine can be asymptomatic or is associated with axial pain, radicular pain, or neurologic impairment, depending on the size and location of the tumor [13]. In our case, the occasional photopsia was experienced. However, the detail reasons were not clarified, although in fact it disappeared after the resection of tumor.

Findings on film radiographs of patients with GCTTS vary, including circumscribed soft tissue tumors (approximately 50%), negative findings (19.4%), bone erosion (13.9%), periosteal reaction (8.3%), intralesional calcifications (5.5%), and permeative intraosseous invasion (2.9%) [4]. On CT scans, the lesion may demonstrate hyperattenuation resulting from the hemosiderin content [5]. The matrix of the tumor is rarely calcified. Sometimes the lesion occurs as a destructive bone lesion with pressure erosion and sclerotic margins of the posterior elements of the vertebra [14]. On MRI, the lesion is commonly isointense relative to the muscle on T1-weighted images. The variable intensity on T2-weighted images can be found because of the variable content of hemosiderin, liquid, lipids, fibrous tissue, and hemorrhage [6,14]. Positron emission tomography can visualize and differentiate between low- and high-grade soft tissue sarcomas. Uptake in PET correlates with the proliferation of soft tissue sarcomas [15]. Hamada et al. [16] and Shin et al. [17] reported a statistically significant difference in SUVmax between malignant and benign lesions. However, the PET image features of GCTTS have not been previously described in the literature.

In our case, the findings on plain radiographs were negative; CT images revealed a mildly enhanced tumor behind the posterior arch of C1, with no erosion of adjacent bones and no expansile bony lesions. The magnetic resonance characteristics in our case are consistent with the findings described in the literature. However, the magnetic resonance findings are by no means diagnostic of GCTTS, and we initially considered desmoid, fibroma, or other malignancies such as fibrosarcoma or metastatic carcinoma. In addition, on PET, the lesion displayed a high SUV (max: 10.10), resembling a malignant tumor. Therefore, we elected to perform a biopsy to aid in a definitive diagnosis.

Histologic analysis of frozen sections showed a benign-appearing hypercellular tumor with a mixture of cell sizes. Based on the diagnosis, we removed the tumor completely. The primary consideration guiding therapeutic intervention for GCTTS is its tendency to recur locally. Complete resection is curative, and every effort should be made to achieve a gross total excision at the time of the first surgery. In cases where total excision was not possible, early recurrence was noted. In addition, the estimated rate of local recurrence of GCTTS in the spinal region is 18%–25%, which is comparable with that of GCTTS of the appendicular skeleton [18,19]. Repeated surgical resection for recurrent spinal lesions is typically curative, but subtotal excision may provide adequate tumor control. After 3 years of follow-up following marginal en bloc resection, our patient has no signs of local relapse as determined by imaging.

The effectiveness of radiotherapy to prevent recurrence after initial gross total tumor excision is unclear. Radiotherapy should be reserved for inoperable cases, thereby avoiding the potential for radiation-induced neurologic damage and the smaller risk of post-radiation sarcoma [5]. Recent reports describe the use of imatinib for treating GCTTS in patients with recurrent or unresectable lesions [20–22]. Imatinib-induced inhibition of tumor growth is thought to occur through its activity against colony-stimulating factor 1 receptor [22,23].

Although GCTTS of the spine is extremely rare, awareness of this tumor is important because its radiological features may simulate other neoplastic processes in the spine. The tumor in our case presumably arose from the membrane surrounding the posterior arch of the C1 vertebra and was somewhat unusual in that we were unable to detect any association with the adjacent facet joint or the C1 occipital-condyle joint. Only one case similar with ours has been reported. In the previously reported case, the lesion arose not from a facet joint but from a small bursa adjacent to the posterior C1 arch [9]. The majority of GCTTS tumors involve or are centered at the facet joint. However, GCTTS can occur in the site away from the facet joint, which is an important consideration when making the diagnosis of GCTTS of the spine.

Conclusions

We experienced an extremely rare case of GCTTS arising from the membrane surrounding the posterior arch of the C1 vertebra. In spite of the rarity of this disease, GCTTS should

be considered in the differential diagnosis of the axial skeletal lesion. Awareness of GCTTS is important because its radiographic features may simulate other neoplastic lesions in the spine.

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