Tenosynovial Giant Cell Tumor at the Upper Cervical Spine likely Arising from the Posterior Atlantoaxial Membrane: A Great Radiological Mimic

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ABSTRACT

Although tenosynovial giant cell tumor (TSGCT) is commonly found in the limbs along tendon sheaths, bursae, and synovial joints, its occurrence in the axial skeleton is rare. Majority of the cases of TSGCT of the spine reported in the English literature were found to arise from the spinal facet joints. We report a rare case of TSGCT in the upper cervical spine likely arising from the posterior atlantoaxial membrane, complete with detailed magnetic resonance imaging (MRI) and computed tomography (CT) findings, and an approach to CT-guided biopsy of such paraspinal lesions.

CASE REPORT

CASE REPORT

A 54 year-old lady with a history of resected parotid oncocytoma was found to have an incidental right paraspinal enhancing nodule on her surveillance MRI neck study. She had neither constitutional symptoms nor neurological deficits on clinical history and examination. Her laboratory results were normal. She was referred to the orthopedic surgery clinic. Although the patient was asymptomatic, this nodule showed slight increase in size on follow-up MRI study performed a year later. Hence, she was counselled for a biopsy which was performed under CT guidance within a month.

Histopathological analysis showed the neoplasm to be a tenosynovial giant cell tumor (TSGCT). Follow-up MRI study performed two years following the initial MRI study showed the lesion to be stable. Patient remains asymptomatic at three years of follow-up.

Radiological Description

On MR imaging, there was a 1.5 x 0.9 x 1.1 cm well-defined ovoid nodule, lying just posterior to the right posterior arch of C1 vertebra. No underlying bony erosion nor invasion was seen. Medially, it mildly displaced the nuchal ligament to the left. Posteriorly, it gently displaced the right rectus capitis muscle posterolaterally. It demonstrated homogenous isointensity to the adjacent muscles on T1- and T2-weighted sequences. Some susceptibility artifacts were seen within the lesion on the gradient echo sequence, suggestive of hemosiderin deposition. It demonstrated homogenous restricted diffusion, and heterogeneous but avid contrast enhancement. No extension into the spinal canal or neural foramina was detected. It appeared localized and showed a tail with the posterior atlantoaxial membrane (Figure 1). On unenhanced CT imaging, it was slightly hypodense to the adjacent paraspinal muscles, with an average Hounsfield unit of 35 (Figure 2).

CT-Guided Biopsy Procedure

The procedure was performed under local anesthesia without conscious sedation. The patient was positioned prone, with the head facing left. Pre-biopsy on-table CT showed the nodule to be stable in size and appearance. The entry site was planned to be at the left posterolateral neck, in order to achieve a good length of core biopsy tissue along the long axis of the nodule. 1% lignocaine was given as local anesthesia. An 18G x 16 cm Bard Mission biopsy needle was advanced into the nodule and 3 good cores of tissue were obtained. Post-biopsy CT showed no hematoma. The patient was discharged well on the same day.

Pathological Description

The section showed cores of lesional tissue featuring a proliferation of bland mononuclear cells surrounded by fibrosis.

The cells had uniform round to ovoid nuclei and inconspicuous nucleoli with scant to moderate amounts of light eosinophilic cytoplasm. Some of the cells contained eccentrically located nuclei and a peripheral hemosiderin rim. Aggregates of foamy and hemosiderin-laden macrophages and scattered multinucleated giant cells were seen (Figure 3). There was no marked cytological atypia or necrosis. The above findings may be consistent with a tenosynovial giant cell tumor, which can rarely occur in the spine. The distinction between localized versus diffuse type could not be determined on the biopsy.

DISCUSSION

Etiology & Demographics

Tenosynovial giant cell tumor (TSGCT) is a benign fibrous histiocytic tumor arising from the synovium of tendon sheaths, bursae and joints [1]. It is commonly found in the upper and lower extremities, such as in the hands and knees. These tumors are usually seen in the third and fourth decades of life. However, the occurrence of TSGCT in the axial skeleton is rare, with less than a hundred cases reported in the English literature. The first report of TSGCT of the spine was in 1980 by Kleinman et al. [2]. This poses a diagnostic challenge to radiologists as it may mimic other neoplasms of the spine such as meningioma, chordoma, and malignant conditions such as metastasis.

TSGCTs are classified into two types: diffuse or localized. Localized-type TSGCT primarily occurs in the fingers and demonstrates distinct margins, whereas diffuse-type TSGCT occurs in larger joints such as the hip, knee, shoulder, and lacks defined margins without encapsulation. Diffuse-type TSGCTs are also called pigmented villonodular synovitis (PVNS). In the literature, some TSGCTs in the spine are thought to arise from the synovium of spinal accessory joints such as the facet joint. A systematic review of TSGCTs in the spine showed that 77.1% of these spinal TSGCTs were found the cervical and lumbar spine, and they occur more often in women than in men [3]. Apart from our case, we found only 2 other cases of TSGCT arising from the posterior atlantoaxial membrane in the English literature [4,5].

Clinical and Imaging Findings

Amongst the few reported cases in the English literature, some cases were symptomatic with neck or back pain, radiculopathy, numbness and weakness cited as the more common presenting symptoms [6]. Some cases, as with our case, were asymptomatic with completely normal neurological examination. These cases were often detected as incidental findings on imaging.

On imaging, a comprehensive review of the literature showed majority of the tumors originating from the facet joint and showing expansion of tumor into the spinal canal with bony erosions [3,6]. However, their signal characteristics on MRI vary amongst reports. On T1-weighted sequences, some TSGCTs show isointense signal to muscle, while others are heterogeneous in signal. On T2-weighted sequences, there is a range of reports from homogeneous hypo- and hyperintense, to heterogeneous signals. However, most cases demonstrate avid contrast enhancement with Gadolinium. Some cases demonstrate susceptibility artifacts on gradient echo sequences suggestive of hemosiderin deposition. This imaging characteristic is similar to their counterpart commonly found in the limbs and has been described as pathognomonic to suggest a diagnosis of TSGCT, but the absence of it does not exclude a TSGCT [7, 8].

Differential Diagnoses

The imaging features of spinal TSGCTs are often varied and non-specific, due to the variable proportion of hemosiderin, lipid, fibrous tissue, cystic and cellular components within [9]. Like most soft tissue tumors, TSGCT shows moderate to marked enhancement following contrast administration [9,10]. Its atypical location in the spine further contributes to the diagnostic challenge, making it prone to being a great radiological mimic of other commonly thought of spinal neoplasms such as schwannoma, meningioma, neurofibroma, chordoma, fibroma of tendon sheath, hemangioma, and the most feared - metastasis.

A study by Lee JH reported that schwannomas often showed cystic change, neural foraminal extension, and are lumbar in location, while meningiomas showed higher frequency of dural tail sign and thoracic location [11]. Calcifications are more often found in meningiomas, whereas schwannomas do not typically calcify [12]. Spinal TSGCTs do not typically show dural tail sign, and susceptibility artifacts within spinal TSGCTs, if present, are due to hemosiderin deposits. Meningioma is typically isointense to grey matter on T1-weighted sequences, isointense or hyperintense to grey matter on T2- weighted sequences, and demonstrates intense contrast enhancement. Schwannoma is often isointense or hypointense on T1-weighted sequences, heterogeneous to hyperintense on T2- weighted sequences, and demonstrates intense contrast enhancement. Target sign may be observed in schwannomas (peripheral T2-weighted hyperintensity, with central T2-weighted hypointensity).

Neurofibroma and plexiform neurofibroma are typically T1-weighted hypointense, T2-weighted hyperintense, and show heterogeneous contrast enhancement. A known personal or familial history of neurofibromatosis would aid in the diagnosis; however, the majority are sporadic. Fascicular sign is commonly observed in neurofibroma, while calcification is uncommon [13].

Chordoma is a low-grade malignant neoplasm arising from notochord remnants. The cervical spine is affected in 6% of cases and extra-axial chordomas are very rare. It is typically seen as an osteolytic soft tissue mass in the midline [14]. They are usually T1-weighted hypointense to isointense, markedly T2-weighted hyperintense due to high fluid content, and show moderate contrast enhancement [15].

Fibroma of tendon sheath (FTS) is a benign tumor arising from the synovium of the tendon sheath. Most FTS occurs around small joints such as the fingers, hands, and wrist [16].

FTS is rarely seen around a large joint, let alone in the spine. Similar to TSGCT, FTS are often hypointense on T1- and T2-weighted sequences. However, FTS shows mild or lack of contrast enhancement, whereas TSGCT shows more prominent contrast enhancement [16].

Extramedullary hemangioma in the spine is rare but could mimic TSGCT due to its low signal intensity on T1-weighted sequence, and the presence of susceptibility artifacts on T2* sequence (due to phleboliths). However, hemangiomas often demonstrate T2-weighted hyperintensity and avid contrast enhancement. The presence of signal voids on T2-weighted sequences will also raise the suspicion of a lesion of vascular origin. [17]

Paraspinal metastasis is often multiple, in a random distribution, and within the paraspinal muscles, rather than in close association with a facet joint or along the ligaments of the spine. Metastasis will also show imaging features similar to their primary malignant tumor.

In conclusion, the periarticular or perimembranous location of the tumor, the presence of avid contrast enhancement, the presence of susceptibility artifacts within suggesting hemosiderin deposits, and the absence of a dural tail, will be useful imaging characteristics to guide a radiologist to consider the diagnosis of a TSGCT despite its atypical location in the spine.

Treatment & Prognosis

In view of the variable imaging features of spinal TSGCTs, most patients eventually require a biopsy for definitive diagnosis. Depending on the tumor's location, this may be performed via endoscopy or under CT guidance.

Poutoglidis et al reported a case of TSGCT in the posterior pharyngeal space, just anterior to the left lateral mass of C1, which was mobilized under endoscopic assistance and excised en-bloc [18].

For paraspinal tumors in the lateral or posterior location, CT-guided biopsy remains the safest option. We suggest positioning the patient in prone position, with the head facing the contralateral side (i.e. away from the tumor). This will allow a longer trajectory for the biopsy needle to be stabilized within the paraspinal muscles. As much as possible, the biopsy should be performed along the long axis of the tumor, to obtain as much tissue as possible within a single core.

For asymptomatic patients, such as in our case, spinal TSGCTs are managed conservatively. However, if there are symptoms affecting daily living due to mass effect by the tumor, gross total resection (GTR) was the best treatment strategy [3]. The recurrence rate of patients who underwent GTR was 7.7%, and tumor progression was observed in 66.7% of patients who underwent subtotal resection [3].

TEACHING POINT

TSGCT arising from the posterior atlantoaxial membrane is an extremely rare entity in which the periarticular or perimembranous location of the tumor, the presence of avid contrast enhancement, the presence of susceptibility artifacts suggesting hemosiderin deposits, and the absence of a dural tail, will be useful imaging characteristics to guide a radiologist to consider its diagnosis despite its atypical location in the spine. TSGCTs located just posterior to the nasopharynx are preferably biopsied or excised via the endoscopic approach, whereas TSGCTs located along the posterior or lateral spine are better approached under CT guidance with the patient in prone position and head facing away from the tumor.

QUESTIONS

QUESTION 1: Which of the following answer choices is INCORRECT regarding spinal tenosynovial giant cell tumor?

1. Majority of spinal tenosynovial giant cell tumors (TSGCTs) occur in the neck and lower back.
2. Spinal TSGCTs are more often found in men than women. (applies)
3. Most spinal TSGCTs arise from the spinal facet joints.
4. TSGCTs are broadly classified into diffuse and localized types.
5. Spinal TSGCTs can cause bony erosion.

Explanation:

1. Most spinal TSGCTs are found in the cervical and lumbar spine. [A systematic review showed that... 77.1% of these spinal TSGCTs were found the cervical and lumbar spine.]

2. Amongst the cases reported in the English literature, more women than man are affected. [A systematic review showed that... they occur more often in women than in men.]

3. The most common site of origin of spinal TSGCT is the facet joint. [A comprehensive review of the literature showed majority of the tumors originating from the facet joint.]

4. There are two types of TSGCTs - diffuse and localized. [Localized-type TSGCT primarily occurs in the fingers and demonstrates distinct margins, whereas diffuse-type TSGCT occurs in larger joints... and lacks defined margins without encapsulation.]

5. Bony erosions are common in spinal TSGCTs. [A comprehensive review of the literature showed majority of the tumors... showing expansion of tumor into the spinal canal with bony erosions.]

QUESTION 2: Regarding imaging findings of spinal TSGCTs, which of the following answer choice is INCORRECT?

1. Blooming artifacts may be observed.

2. Contrast enhancement is seen.
3. T1- weighted hyperintensity is always demonstrated. (applies)
4. Spinal TSGCT can be found in close association with a joint or ligament.
5. There is absence of a dural tail.

Explanation:

1. Spinal TSGCT may contain hemosiderin. [Some susceptibility artifacts were seen within the lesion on the gradient echo sequence, suggestive of hemosiderin deposition.]

2. Spinal TSGCT enhances avidly. [TSGCT shows moderate to marked enhancement following contrast administration]

3. T1 signal varies from hypointense to heterogeneous. [On T1-weighted sequences, some TSGCTs show isointense signal to muscle, while others are heterogeneous in signal]

4. Spinal TSGCT can arise from facet joint or membrane. [The periarticular or perimembranous location of the tumor... will be useful imaging characteristics to guide a radiologist to consider the diagnosis of a TSGCT.]

5. Dural tail is an imaging feature of meningiomas. [Meningiomas showed higher frequency of dural tail sign and thoracic location.]

QUESTION 3: All of the following could be radiological mimics of spinal TSGCT except:

1. Meningioma
2. Schwannoma
3. Extramedullary hemangioma
4. Chordoma
5. Hepatocellular carcinoma (applies)

Explanation:

1. Both spinal TSGCT and meningioma may show susceptibility artifacts [Calcifications are more often found in meningiomas... Susceptibility artifacts within spinal TSGCTs are due to hemosiderin deposits.]

2. Both spinal TSGCT and schwanomma demonstrate similar T1- and T2- weighted signal characteristics, and show avid contrast enhancement. [Schwannoma is often T1- weighted isointense or hypointense, T2- weighted heterogeneous to hyperintense, and also demonstrate intense contrast enhancement.]

3. Both spinal TSGCT and extramedullary hemangiomas enhance avidly and may show susceptibility artifacts. [Extramedullary hemangioma in the spine is rare but could mimic TSGCT due to... the presence of susceptibility artifacts... and avid contrast enhancement.]

4. Both spinal TSGCT and chordoma demonstrate similar T1-weighted signal characteristics and show contrast enhancement. [Chordomas... are usually T1- weighted hypointense to isointense... and show moderate contrast enhancement]

5. Hepatocellular carcinoma is found in the liver.

QUESTION 4: Which of the following is not a main consideration when performing biopsy of a paraspinal tumor?

1. Patient position
2. Shape of the tumor
3. Location of the tumor
4. T1-weighted signal characteristics (applies)
5. Mode of biopsy

Explanation:

1. The patient is often position supine for endoscopic guided biopsy, and may be positioned prone for CT-guided biopsy depending on the exact location of the tumor. [For paraspinal tumors in the lateral or posterior location, CT-guided biopsy remains the safest option. We suggest positioning the patient in prone position...]

2. Biopsy should be performed along the longest axis of the tumor to obtain a good sample. [As much as possible, the biopsy should be performed along the long axis of the tumor, to obtain as much tissue as possible within a single core.]

3. Tumors located just posterior to the nasopharynx could be biopsied via endoscopic approach. [Poutoglidis et al reported a case of TSGCT in the posterior pharyngeal space...which was mobilized under endoscopic assistance and excised en-bloc.]

4. T1 signal characteristics are observed on MRI. Biopsy is often performed under CT rather than MRI guidance. [For paraspinal tumors... CT-guided biopsy remains the safest option.]

5. Biopsy of paraspinal tumor may be performed under CT or endoscopic guidance. [TSGCTs located just posterior to the nasopharynx are preferably biopsied or excised via the endoscopic approach, whereas TSGCTs located along the posterior or lateral spine are better approached under CT guidance with the patient in prone position and head facing away from the tumor.]

QUESTION 5: Which of the following is not a symptom associated with spinal TSGCT?

1. Hematochezia (applies)
2. Neck or back pain
3. Radiculopathy
4. Numbness
5. Weakness

Explanation:

1. Hematochezia is seen in conditions resulting in bleeding within the gastrointestinal tract.

2. Depending on its location in the spine, TSGCT may cause pain in the neck or back. [some cases were symptomatic with neck or back pain]

3. Shooting pain down the limbs is noted in some cases of spinal TSGCT. [some cases were symptomatic with... radiculopathy]

4. Depending on the site of compression on the spinal cord or nerve root, spinal TSGCT may cause numbness. [numbness and weakness cited as the more common presenting symptoms]

5. Depending on the site of compression on the spinal cord or nerve root, spinal TSGCT may cause weakness. [numbness and weakness cited as the more common presenting symptoms]

AUTHORS' CONTRIBUTIONS

YEL, conceptualization, clinical data, writing and editing
CHMQ, interpretation, writing and editing
YLT, interpretation, pathological data, writing and editing
AP, interpretation and editing
JAY, interpretation, writing and editing, final approval of manuscript

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DISCLOSURES

Nil

CONSENT

Yes. We have obtained informed written consent from the patient.

HUMAN AND ANIMAL RIGHTS

Nil

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FIGURES

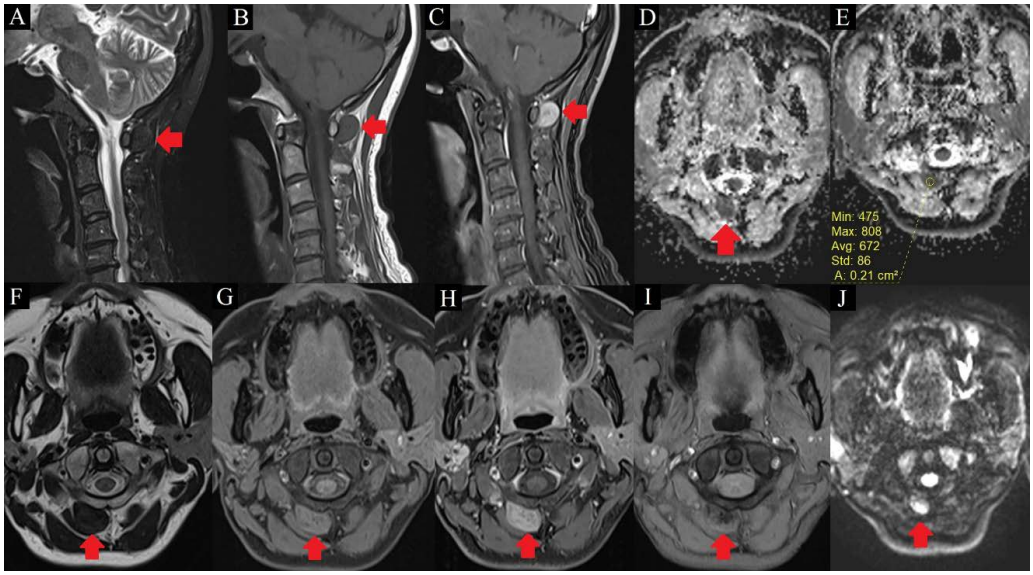

Figure 1: MRI images of a 54 year-old lady with tenosynovial giant cell tumor at the upper cervical spine, likely arising from the posterior atlantoaxial membrane.

MRI specifics: Siemens, 1.5 Tesla, 10mL Clariscan as intravenous contrast media.

Subfigures: (A) Sagittal T2 STIR, (B) Sagittal T1 pre-contrast, (C) Sagittal T1 fat-saturated post-contrast, (D) Axial ADC, (E) Axial ADC value, (F) Axial T2, (G) Axial T1 fat-saturated pre-contrast, (H) Axial T1 fat-saturated post-contrast, (I) Axial GRE, (J) Axial DWI

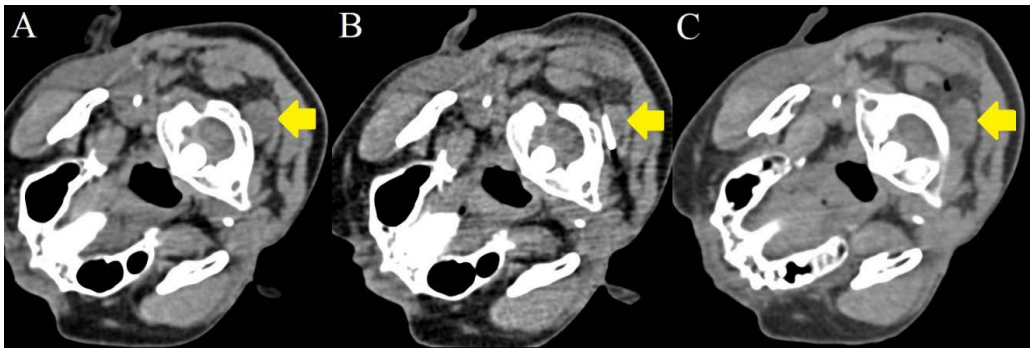

Figure 2: CT images of a 54 year-old lady with tenosynovial giant cell tumor at the upper cervical spine, likely arising from the posterior atlantoaxial membrane.

CT specifics: Siemens, Slice thickness 1.5mm, 120kV. (Soft tissue window)

Subfigures: (A) Axial pre-biopsy, (B) Axial during biopsy (needle within tumor), (C) Axial post-biopsy (gas seen along biopsy tract)

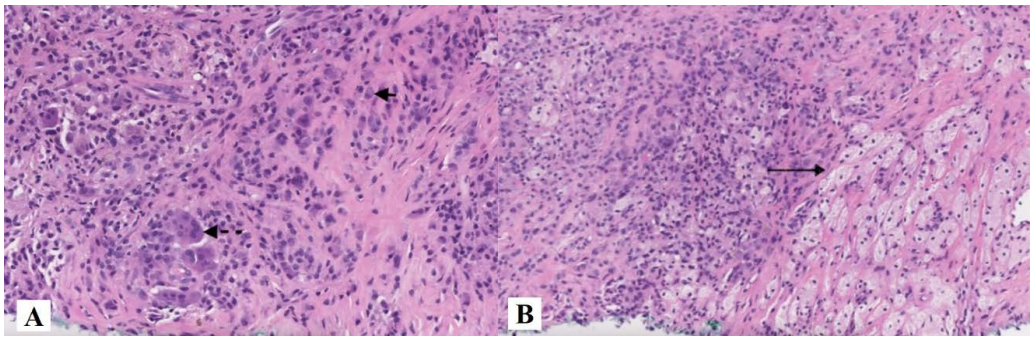

Figure 3: (A) Microscopic findings revealed the tumor comprises a mixture of mononuclear cells (solid black arrow), some containing a peripheral rim of hemosiderin, and scattered multinucleated giant cells (dashed black arrow) in a background of fibrosis. (B) Foamy macrophages are associated with the tumor (long black arrow).

TABLES

Table 1: Summary table of tenosynovial giant cell tumor in the spine

Etiology	Thought to arise from synovial membrane (most commonly of the spinal facet joints)
Incidence	77.1% of spinal TSGCTs were found the cervical and lumbar spine. Only 2 other cases of TSGCT have been found to arise from the posterior atlantoaxial membrane in the English literature
Gender ratio	More often in women than in men
Age predilection	Third and fourth decades of life
Risk factors	Unknown
Treatment	Surveillance if asymptomatic. Gross total resection if symptomatic.
Prognosis	Low recurrence rate (7.7%) following gross total resection.
Findings on imaging	T1 isointense to muscle, T2 variable, presence of susceptibility artifacts and restricted diffusion, avid heterogeneous contrast enhancement

Table 2: Differential diagnoses and their imaging findings

Differential Diagnoses	**T1-weighted MRI**	**T2-weighted MRI**	**T2* MRI**	**Contrast enhancement**	**Distinguishing features**
Schwannoma	Isointense or hypointense	Heterogeneous Hyperintense Target sign	Calcification is uncommon	Heterogeneous Intense	Cystic changes Neural foraminal extension More commonly lumbar in location
Meningioma	Isointense to grey matter	Isointense or hyperintense to grey matter	Susceptibility artifacts due to calcifications	Intense	Dural tail sign More commonly thoracic in location
Neurofibroma	Hypointense	Hyperintense Fascicular sign	Calcification is uncommon	Heterogeneous	May be associated with NF1/2
Chordoma	Hypointense to isointense	Markedly hyperintense	Susceptibility artifacts due to calcifications and/or hemorrhage	Moderate	Often found midline in location
Fibroma of tendon sheath	Hypointense	Hypointense	Calcification is uncommon	Mild or none	Often found around small joints
Extramedullary hemangioma	Hypointense	Hyperintense	Susceptibility artifacts due to phleboliths	Intense	Signal voids

KEYWORDS

Tenosynovial giant cell tumor; Giant cell tumor of the tendon sheath; Pigmented villonodular synovitis; Posterior atlantoaxial membrane; Spine

ABBREVIATIONS

ADC = Apparent Diffusion Coefficient
CT = Computed Tomography
DWI = Diffusion Weighted Imaging
FTS = Fibroma of Tendon Sheath
GRE = Gradient Echo
GTR = Gross Total Resection
MRI = Magnetic Resonance Imaging
PVNS = Pigmented Villonodular Synovitis
STIR = Short Tau Inversion Recovery
TSGCT = Tenosynovial Giant Cell Tumor

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