Extradural cervical spinal meningioma mimicking malignancy

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ABSTRACT

The vast majority of intraspinal meningiomas occur in an intradural extramedullary location. A meningioma in a purely extradural location in the cervical spine as reported here is quite exceptional. Extradural meningiomas tend to show more aggressive features than intradural meningiomas and are often confused with malignant neoplasms. We report an invasive extradural meningioma in the cervical spine with multi-segmental involvement, extension through the neural foramina and encasement of the subjacent vertebral artery, mimicking malignancies such as lymphoma and sarcoma. Although rare, meningiomas may demonstrate extradural multi-segmental growth and should be considered in the differential diagnosis of such lesions.

CASE REPORT

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A previously well 35 year old Chinese male presented with insidious onset of neck pain, numbness and stiffness in all four limbs and bilateral upper limb and truncal loss of temperature sensation over several months. Neurological examination revealed mildly reduced power in all four limbs with spastic tone and gait. Upper and lower limb reflexes were brisk and ankle clonus was present. Plantar responses were upgoing bilaterally. There was decreased temperature sensation from T4 to T12 dermatomes, decreased vibration sensation from C6 to T1 dermatomes and decreased proprioception in bilateral upper limbs. The rest of the neurological examination was normal. Blood workup was unremarkable.

Magnetic resonance imaging (MRI) of the cervical spine (Figure 1 and Figure 2) showed an infiltrative, extradural soft tissue lesion within the right side of the spinal canal extending from C1 to C4. This lesion was isointense to cervical cord on T1-weighted (T1W) and T2-weighted (T2W) sequences, with uniform postcontrast enhancement. No restricted diffusion was seen. The lesion extended through the expanded right neural foramina into the surrounding muscle planes of the deep upper neck. The right vertebral artery was encased at the transverse foramina, but remained patent. At C3 level, there was cord compression with intramedullary T2W hyperintensity suggesting cord edema.

Computed tomography (CT) angiogram (Figure 3) demonstrated widening and scalloping of the affected C1 to C4 neural foramina but no bony destruction. The right vertebral artery remained normal in caliber despite encasement by the lesion.

Based on imaging findings, the lesion was thought to represent either lymphoma or plexiform neurofibroma. Lymphoma was considered because of the lesion's T2W isointense signal, which is unusual for most other extradural tumors which tend to be T2W hyperintense. Encasement without stenosis or invasion of the adjacent vessel also favored a diagnosis of lymphoma. Another differential considered was

that of a plexiform neurofibroma. Although our patient did not have any known history or stigmata of neurofibromatosis, the extension of tumor through the neural foramina with bony scalloping on CT was suggestive of a neurogenic tumor. Soft tissue sarcoma and metastasis were less favored as bony scalloping of the affected neural foramina favored a slow growing lesion and there was no evidence of mineralized matrix or bony destruction on CT.

Anterior decompression and fusion of the cervical spine with C2 and C3 total laminectomy, C1 and C4 partial laminectomy and fusion of C2 and C3 was performed. Only subtotal resection of the lesion was performed due to its encasement of the right vertebral artery and lateral invasion into the surrounding soft tissue.

Histopathology revealed meningothelial meningioma (WHO grade I) with invasion of fat planes and fibrous tissue (Figure 4).

Post-operative course was uneventful. There was complete resolution of truncal numbness and power in all limbs returned to full.

Postsurgical follow up MRI (Figure 5) showed decompression of the cervical cord with residual tumor encasing the patent right vertebral artery and residual lateral soft tissue extension. Continued follow up for three years did not reveal any increase in dimension of residual tumor.

DISCUSSION

Etiology & Demographics:

Meningiomas are common intraspinal lesions, accounting for 25 – 46% of primary intraspinal neoplasms [1]. These are typically intradural extramedullary in location, being most likely derived from the meningothelial cells of the arachnoid layer. Extradural spinal meningiomas are infrequent (2.5 to 3.5% of all spinal meningiomas) [2], while exclusively extradural meningiomas are very rare [1,3,4].

There are several hypotheses concerning the extradural growth of meningiomas. One is that meningiomas originate from the nerve root segment where the arachnoid mater contacts the dura mater [5,6]. A second hypothesis is that the tumor originates from aberrant arachnoid islets in the epidural space. This hypothesis is supported by several case reports, such as that of intraorbital meningioma with no relation to the optic nerve sheath, or extracranial meningioma between the galea and the bone [6]. Another hypothesis is that epidural meningioma originates directly from the external surface of the dura mater (cap cells) and grows into the extradural space with a globular or en plaque pattern [7]. This hypothesis is supported by the fact that the outer dural layer is composed of periosteum and not true dura mater, and hence does not contain arachnoidal rests (ie, embryonic meningothelial cells in the arachnoid layer that have an ectopic location in any area outside the dura) [8].

The mean age of diagnosis of extradural spinal meningioma is 38 years (47% younger than 30 years) [9], occurring at younger ages compared with intradural spinal meningiomas which occur mostly in individuals in their 50s and 60s. Of patients with extradural spinal meningiomas, 64.7% are women. Extradural meningiomas are most commonly seen in the thoracic (80%), cervical (16%) and then lumbar (4%) spine [10].

Clinical & Imaging findings:

The symptomatology of meningiomas includes neurological signs and symptoms due to compression of adjacent structures; specific deficits depend on tumor location. Patients commonly experience pain, followed by weakness and sensory and motor changes. Sphincter dysfunction represents a late finding [11]. There is typically a delay between onset of symptoms and diagnosis.

In contrast with their more common intradural counterparts, extradural meningiomas tend to demonstrate a more aggressive growth pattern. More rapid clinical course has previously been reported, with patients progressing from intact motor function to paraplegia in four weeks [12]. On imaging, they also tend to show more invasive features, including infiltration into the surrounding soft tissues and bony destruction of the adjacent vertebral arch and pedicle.

Some authors have postulated that the appearance of meningiomas in the extradural space of the spinal canal may be a manifestation of the natural tendency of these lesions to invade dura. The frequent invasion of dural sinuses by meningiomas is well known. Hassin showed that the arachnoid villi in the spinal canal occur predominantly at the nerve root exits, and are hence favored "seats of origin" for meningiomas, but leave little intradural space for tumor expansion. Invasion of the dura and extradural tissues thus occurs [13]. This may explain the more rapid time course of these patients, earlier clinical presentation and more invasive appearance of these lesions.

There are many causes of epidural lesions in adults and determination of the cause on the basis of imaging findings and clinical presentation is crucial when deciding to treat a lesion conservatively or with surgery. MR imaging enables further characterization of these lesions. It also accurately demarcates the extent of the lesion and its relation with the spinal cord, providing invaluable information for preoperative planning.

On MRI, spinal meningiomas typically demonstrate T1W isointense signal with iso or hypointense signal on T2W. Contrast enhancement is immediate and homogeneous. This is in contrast to most T2W hyperintense extradural tumors, barring lymphomas which can be T2W hypointense in up to 50% [11]. Restricted diffusion may also be seen in atypical meningiomas.

On CT, calcification in tumors may support a diagnosis of meningioma. However, calcification is visible on neuroimaging in only 1.0%–4.6% of all spinal meningiomas [14]

Differential Diagnoses:

Extradural meningiomas are apt to be confused with other malignant and benign neoplasms which are much more common in this location, yet this distinction is critical to guide the extent of surgery which will impact patient outcome.

Lymphoma

Spinal lymphomas may be paraspinal, vertebral or epidural [15], either in isolation or in combination. Bone marrow involvement indicates stage IV disease. Primary spinal epidural lymphoma with no other recognizable site of disease at the time of diagnosis is much less common [16].

MRI of the whole spine and brain are recommended to determine the complete extent of the disease. The epidural soft tissue appears T1W isointense and T2W iso to slightly hyperintense with intense homogenous enhancement. Up to 50% of lymphomas may show homogeneous T2W hypointense signal intensity due to its dense cellularity. Lymphomas demonstrate restricted diffusion and are hence often the initial working diagnosis in patients with atypical meningiomas, which may also demonstrate restricted diffusion.

Epidural spinal lymphoma almost always demonstrates an infiltrative pattern, with multi-segmental growth and extension through the neural foramina. Histologically, primary spinal epidural lymphoma is commonly of non-Hodgkin's type than Hodgkin's type [17].

Metastases

Metastases are the most common vertebral tumors. Metastases can involve the vertebrae, epidural space, leptomeninges or the spinal cord [18]. The posterior elements, especially the pedicles, may be involved. Osteolytic metastases are most often caused by carcinoma of the lung, breast, thyroid, kidney, and colon while osteoblastic metastases are most commonly caused by prostate carcinoma in men and breast cancer in women.

Epidural soft tissue is commonly associated with the destruction of vertebrae and demonstrates T1W hypointense and T2W hyperintense signal with postcontrast enhancement.

Neurogenic tumor

An extradural tumor extending through the neural foramina would also suggest neurogenic tumors such as schwannoma, neurofibroma or plexiform neurofibromatosis. Neurogenic tumors demonstrate T1W hypointense and T2W hyperintense signal. They classically show a “target sign” of peripheral T2W hyperintense and central hypointense signal due to dense central collagenous stroma.

Chondrosarcoma

Chondrosarcoma is the second most common non lymphoproliferative primary malignant tumor of the spine in adults with peak prevalence between 30 and 70 years of age [19]. The thoracic and lumbar spine are most frequently affected.

Chondrosarcomas of the spine usually manifest as a large, calcified mass with bone destruction [20]. Chondroid matrix mineralization is typical and is best demonstrated with CT. The non mineralized portion of the tumor demonstrates low to intermediate signal intensity on T1W and very high signal intensity on T2W due to the high water content of hyaline cartilage.

Pyogenic abscess

Primary abscesses occur after spinal trauma, injections, surgery, or as a complication of vertebral osteomyelitis, while secondary abscesses occur following hematogenous dissemination of microbes present elsewhere to the epidural space [21]. The most common pathogen in both forms is Staphylococcus aureus.

Abscesses extend laterally and vertically in the epidural space and may occasionally involve the whole length of the spinal canal. Restricted diffusion on MRI confirms the presence of a pyogenic abscess [22].

Aggressive hemangioma

Hemangiomas are benign tumors of blood vessels and can be classified as typical, atypical and aggressive. Aggressive hemangiomas demonstrate invasive radiologic features such as extension beyond the vertebral body, cortical destruction and invasion of the epidural and paravertebral spaces. Aggressive hemangioma can occur at any age, with peak prevalence in young adults [23]. Growth of spinal hemangiomas leading to clinical worsening during pregnancy is a well-known phenomenon, due to hormonal changes and vena cava compression with re-routing of blood to the paravertebral, epidural and azygous venous system.

CT scan reveals hypodense expansile vertebral body mass, with soft tissue extension and spinal cord/nerve root compression. Classical "polka-dot" and "corduroy" signs can guide the correct diagnosis. MRI shows a T1W hypointense, T2W hyperintense lesion with avid contrast enhancement [23].

Epidural lipomatosis

Spinal epidural lipomatosis is characterized by pathological accumulation of unencapsulated adipose tissue in the extradural space [24] and occurs with prolonged steroid use, Cushing's syndrome, or morbid obesity. It most commonly occurs in the thoracic region in the dorsal epidural space and may extend laterally into the paraspinal and psoas muscles with scalloping of vertebral bodies. Epidural lipomatosis is characterized by hyperintense signal both on T1W and T2W images, which is suppressed on fat-saturated images.

Treatment & Prognosis:

Meningioma is a surgically treated disease. The major clinical factor determining rate of recurrence is the extent of resection. However, extent of resection is invariably influenced by tumor site, extent of invasion and attachment to vital intraspinal structures. Furthermore, some histological variants are more likely to recur than others, the most useful morphological predictor of recurrence being the overall WHO grade. Benign meningiomas recur in 7–25% of cases, atypical meningiomas recur in 29–52% and anaplastic meningiomas recur in 50–94% [25]. The recurrence rate after surgery for extradural meningiomas has been shown to be about four times

higher than that of intradural meningiomas [26]. This is probably attributable to difficulty in complete removal of tumor at initial surgery due to its location and dural invasion. Some authors have postulated that if complete resection is achieved, there may be no difference in patients' prognoses [12].

Given the high rate of recurrence, some authors have suggested consideration of radiotherapy as an adjunctive treatment after subtotal excision [27]. Radiotherapy can also help control unexcised or recurrent meningioma.

On the other hand, some authors opine that it is better to reserve adjuvant radiation therapy for recurrent, difficult to reach, high grade cases than after first surgery due to the risk of irradiating a functional spinal cord with no proven benefit. Reoperation should be performed in cases of early recurrence followed by radiotherapy [11].

In our case, subtotal resection was appropriate due to encasement of the right vertebral artery by tumor, which precludes total surgical resection. However, follow up with imaging is mandatory to screen for recurrence.

TEACHING POINT

Spinal meningiomas are typically intradural extramedullary lesions, but may rarely be purely extradural. Extradural meningiomas tend to show more aggressive features and may easily be confused with malignant neoplasms which are much more common in this location. The optimal treatment for meningiomas is total resection with wide margins. When radical resection of tumor poses a risk however, subtotal resection can be a wise option. In such cases, follow up imaging is mandatory to detect tumor recurrence.

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FIGURES

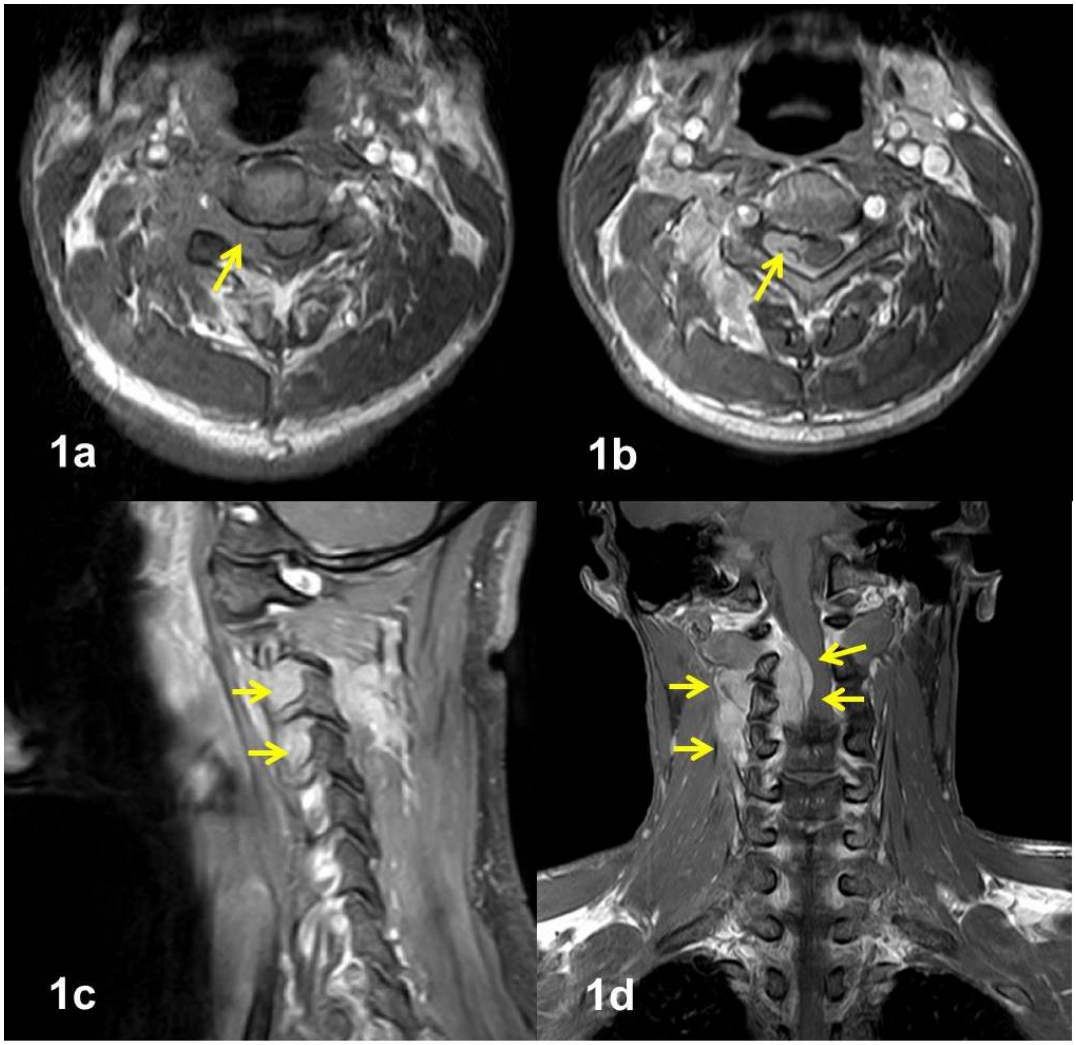

Figure 1: 35 year old man with extradural spinal meningioma.
FINDINGS: MRI of the cervical spine demonstrates a multi-segmental extradural intraspinal mass extending from C1 to C4 levels of the cervical spine. (a) The mass (arrow) is isointense to cervical cord on T1W and (b) demonstrates homogeneous post contrast enhancement. It compresses the cervical spinal cord, (c) extends through the widened C2/3 and C3/4 neural foramina on the right (arrows) and (d) infiltrates laterally into the surrounding muscle planes of the neck.

TECHNIQUE: (a): Axial T1W pre contrast. TR 540. TE 11. 4mm slice thickness. (b): Axial T1W post contrast. 10ml intravenous Dotarem. TR 540. TE 11. 4mm slice thickness. (c): Sagittal T1W post contrast. 10ml intravenous Dotarem. TR 540. TE 11. 4mm slice thickness. (d): Coronal T1W post contrast. 10ml intravenous Dotarem. TR 540. TE 11. 4mm slice thickness.

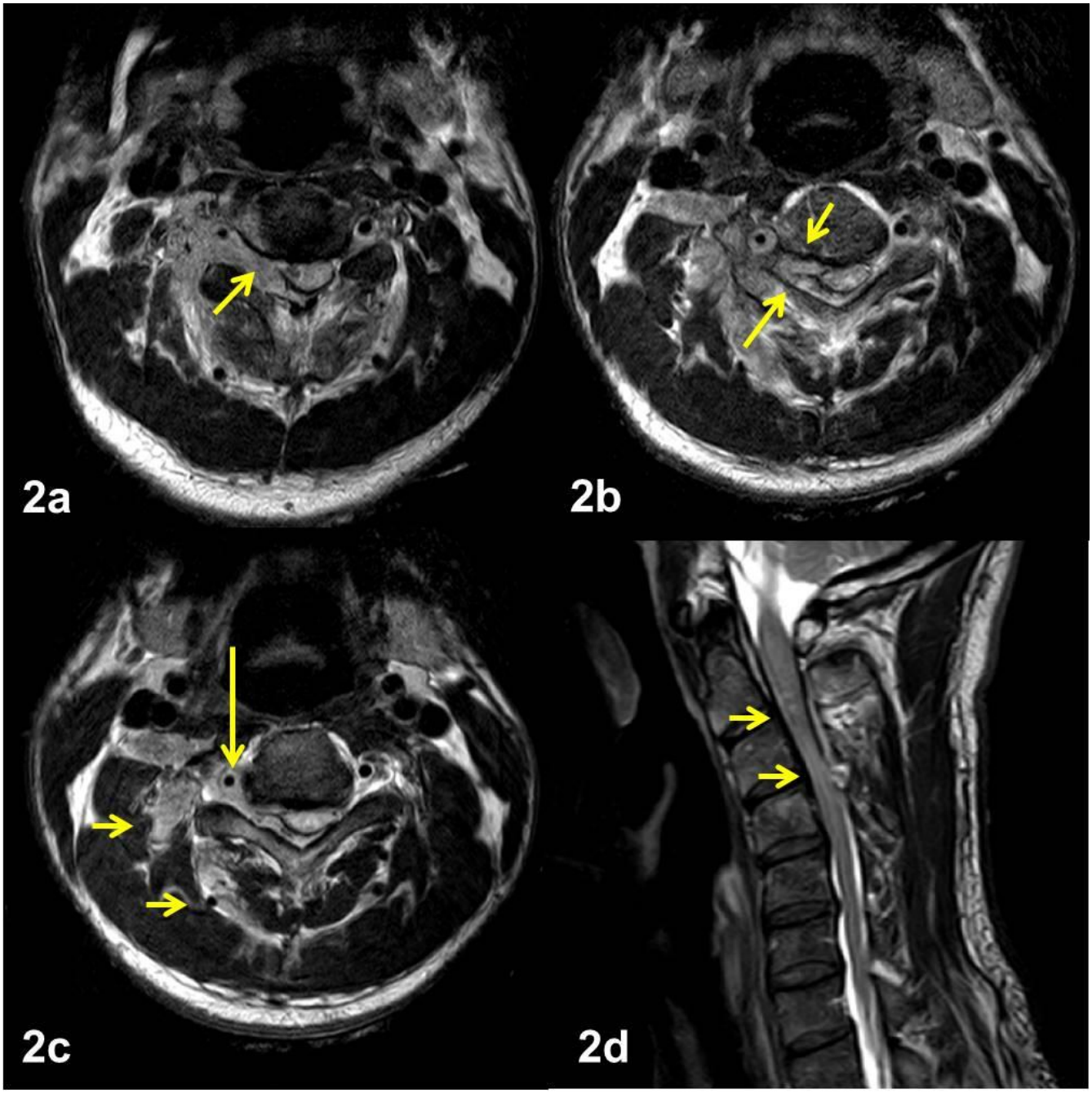

Figure 2: 35 year old man with extradural spinal meningioma.

FINDINGS:
MRI of the cervical spine shows that (a) the intraspinal extradural mass (arrow) is T2W iso to slightly hyperintense to the cervical spinal cord. (b) The mass extends through the widened right neural foramen (arrows) (c) and infiltrates laterally into the surrounding muscle planes of the neck on the right (arrows), in the process encasing the vertebral artery at the right transverse foramen (long arrow). (d) Additionally, the spinal cord demonstrates T2W hyperintense signal from C2 to C4 levels, indicative of cord edema (arrows).

TECHNIQUE:
(a), (b), (c): Axial T2W: TR 4350. TE 105. 4mm slice thickness.
(d): Sagittal T2W: TR 4350. TE 105. 4mm slice thickness.

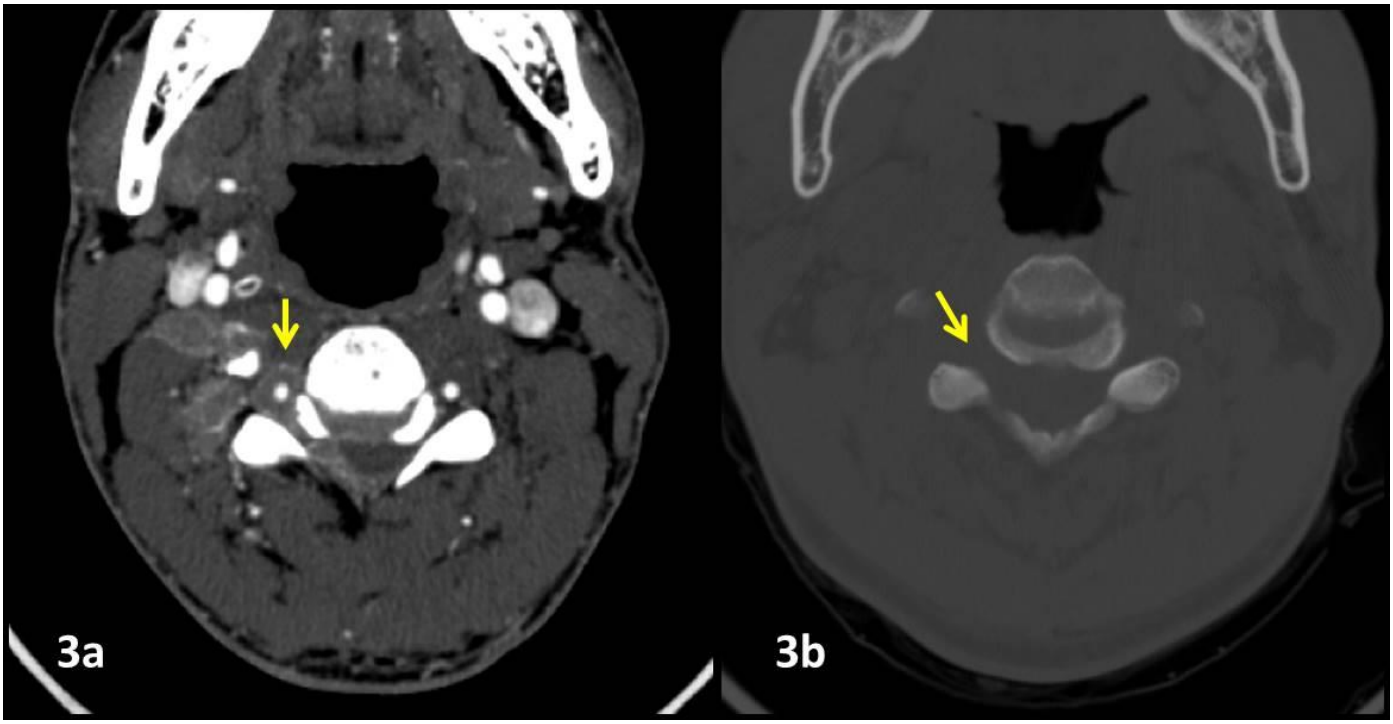

Figure 3: 35 year old man with extradural spinal meningioma.

FINDINGS:
(a) CT angiography of the neck shows that the right vertebral artery (arrow) remains normal in caliber despite encasement by the intraspinal extradual mass. (b) There is widening and scalloping of the neural foramen (arrow) but no bony destruction.

TECHNIQUE:
(a), (b): CT (Toshiba Aquilion) CT angiogram, 350 mAs, 120 kV, 5 mm slice thickness, 60 ml Omnipaque 350

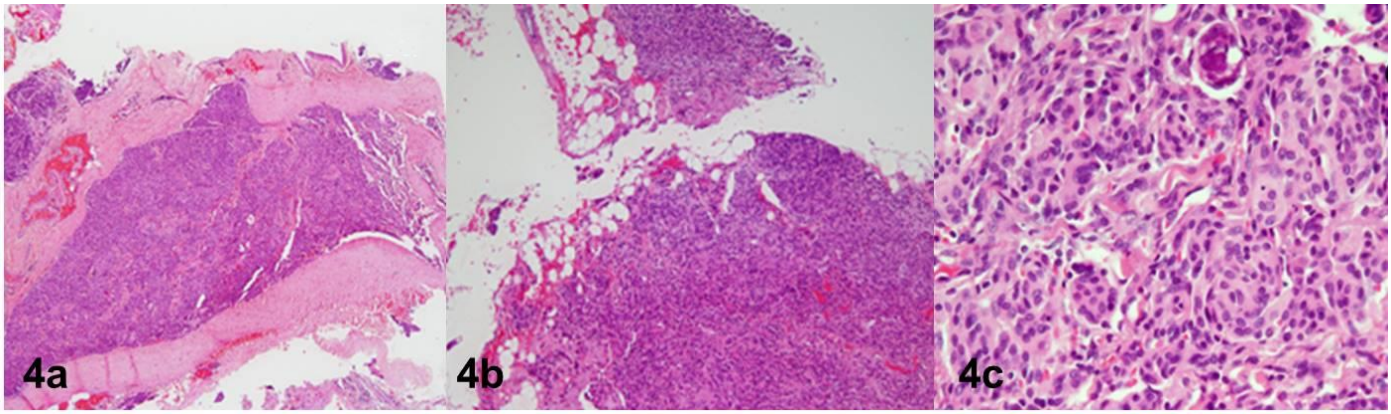

Figure 4: 35 year old man with extradural spinal meningioma.

FINDINGS:
Histopathology paraffin sections from resected cervical spinal extradural lesion show a moderately cellular tumor consisting of fascicles and lobules of meningothelial cells within (a) fibrous dura mater and (b) extradural fibroadipose connective tissue. (c) High power view of the tumor showing fascicles, lobules and whorls of meningothelial cells with uniform ovoid smooth contoured nuclei, delicate eosinophilic cytoplasm and syncytial borders. Psammoma bodies are present.

TECHNIQUE:
(a): Photomicrograph (x40, x100; hematoxylin-eosin stain)
(b): Photomicrograph (x100; hematoxylin-eosin stain)
(c): Photomicrograph (x400; hematoxylin-eosin stain)

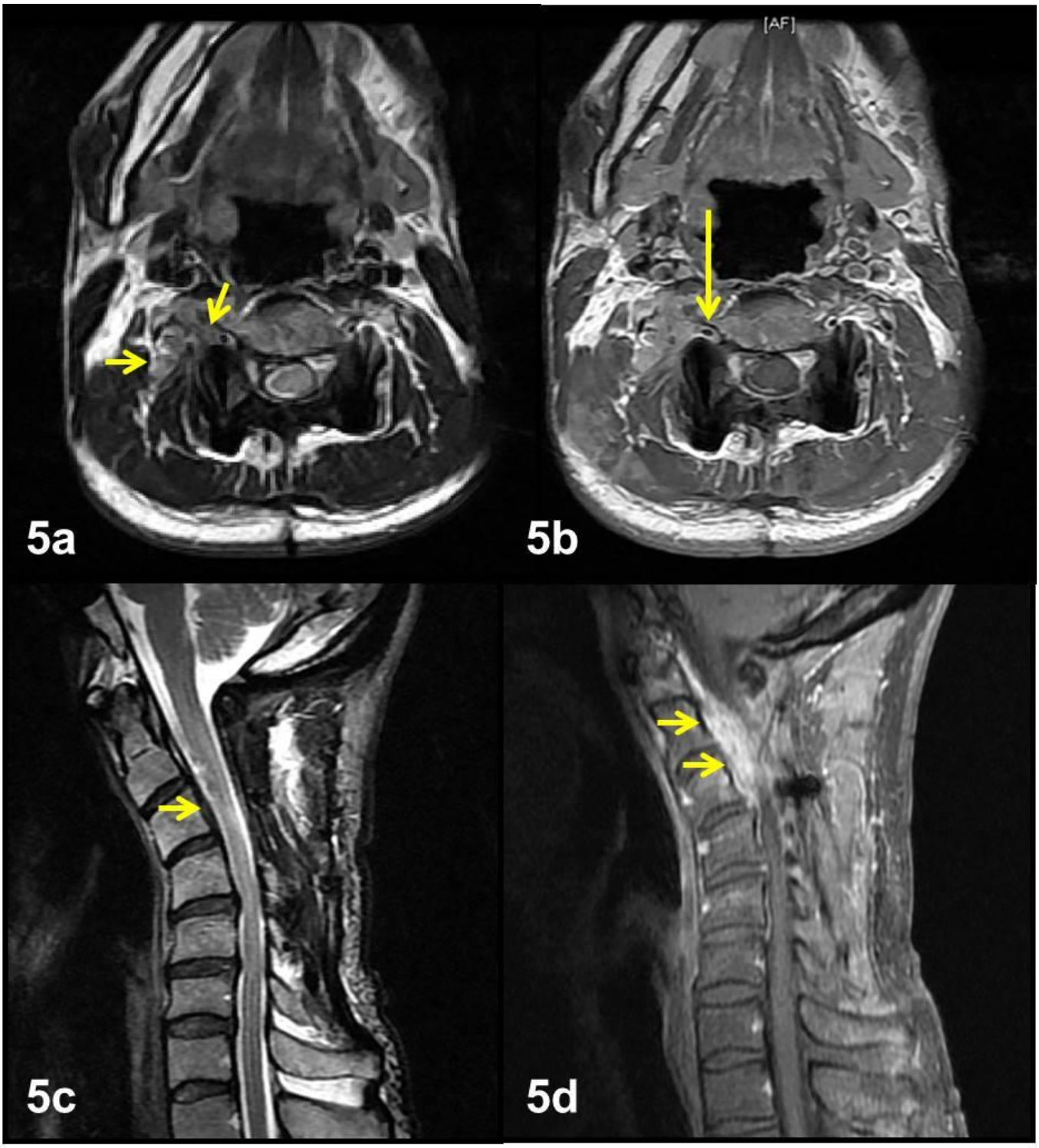

Figure 5: 35 year old man with extradural spinal meningioma, postsurgery.

FINDINGS:
Status post subtotal resection of extradural mass and anterior decompression and fusion of C2 and C3. MRI demonstrates (a, c, d) residual mass (arrows) in the extradural space, infiltrating in between the right neck musculature and encasing the right vertebral artery (long arrow). (b) There is decompression of mass effect on the cervical spinal cord. Residual cervical spinal cord edema at C3 level is noted (arrow).

TECHNIQUE:
(a): Axial T2W: TR 4350. TE 105. 4mm slice thickness.
(b): Axial T1W post contrast. 10ml intravenous Dotarem. TR 540. TE 11. 4mm slice thickness.
(c): Sagittal T2W: TR 4350. TE 105. 4mm slice thickness.
(d): Sagittal T1W post contrast. 10ml intravenous Dotarem. TR 540. TE 11. 4mm slice thickness.

Etiology	Extradural growth pattern is thought to be due to (i) proliferation of ectopic arachnoidal cells around the periradicular nerve root sleeves; (ii) displacement of primitive embryonic remnants of the arachnoid mater and villi along the periradicular dura; or (iii) migration of islands of arachnoid tissue into the extradural space
Incidence	Uncommon, forming 2.5 to 3.5% of all spinal meningiomas. Exclusively extradural meningiomas are rare
Gender Ratio	F > M, 64.7% of patients are female
Age predilection	Occurs at younger ages compared with intradural spinal meningiomas, mean age of diagnosis 38 years (47% of patients younger than 30 years)
Risk Factors	Unknown
Treatment	Total surgical resection. In cases of subtotal resection, follow up imaging is mandatory to assess recurrence. Reoperation should be performed in cases of early recurrence followed by radiotherapy.
Prognosis	Uncertain. Recurrence rate after surgery for extradural meningiomas has been shown to be about four times higher than that of intradural meningiomas.
Findings on imaging	T1W isointense signal, T2W iso or hypointense signal. Contrast enhancement is immediate and homogeneous. Calcification on CT supports diagnosis but is seen in only 1.0%–4.6% of spinal meningiomas.

Table 1: Summary table for extradural spinal meningioma.

Differential diagnosis	**Demographic**	**MRI**	**Helpful distinguishing features**
Extradural meningioma	Mean age at diagnosis 38 years, F >M	T1W iso T2W iso to hypo T1W +C immediate, homogeneous enhancement DWI/ADC Atypical meningiomas may show restricted diffusion	Calcification on CT supports diagnosis but is seen in only 1.0%–4.6% of spinal meningiomas
Lymphoma	40-70, M>F	T1W iso T2W iso to hypo T1W +C avid homogeneous enhancement DWI/ADC Restricted diffusion	Up to 50% may show homogeneous T2W hypointense signal due to its dense cellularity May also show leptomeningeal or intramedullary lesions or bone marrow changes
Metastases	None	T1W hypo T2W hyper T1W +C heterogeneous enhancement	Associated with vertebral destruction CT typically shows sclerotic and/or lytic changes with involvement of the posterior elements
Neurogenic tumour (schwannoma, neurofibroma)	20-30, M=F	T1W hypo T2W hyper T1W +C heterogeneous enhancement	T2W "target sign" due to dense central collagenous stroma Classically shows foraminal extension with "dumbbell" appearance
Chondrosarcoma	30-70, M>F	T1W hypo T2W very hyper T1W +C ring and arc pattern of enhancement	Chondroid matrix mineralization best demonstrated with CT Very high T2W signal intensity due to high water content of hyaline cartilage
Pyogenic abscess	None Risk factors: Diabetes mellitus, spinal intervention, systemic infection e.g. IVDU	T1W hypo T2W hyper T1W +C rim enhancement, no central enhancement	Restricted diffusion on MRI
Aggressive hemangioma	40-60, F>M	T1W hypo to hyper T2W hyper T1W +C avid enhancement	CT "polka dot" or "corduroy" sign due to thickened vertebral trabeculae T2W bright signal due to high water content
Epidural lipomatosis	Risk factors: prolonged steroid use, Cushing's syndrome, morbid obesity	T1W hyper T2W hyper T1W +C no enhancement	Suppression of signal on fat-saturated images

Table 2: Differential diagnoses table for extradural spinal meningioma.

ABBREVIATIONS

CT = Computed tomography
MRI = Magnetic resonance imaging
T1W = T1-weighted MRI sequence
T2W = T2-weighted MRI sequence
T1W +C = T1-weighted post-contrast MRI sequence

KEYWORDS

Extradural; meningioma; spine; intradural; magnetic resonance imaging