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Spinal extradural meningioma en plaque with nerve root attachment and extracanal (intrathoracic) extension. Review of literature on management and case report

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ABSTRACT

Meningiomas are relatively common primary spinal tumours, being the second most common intraspinal tumours probably after vertebral haemangioma.
It constitutes about 25% of all intraspinal tumours; however, in the presence of extradural spinal lesions, the diagnosis of meningioma is uncommon and often not among the first two considerations. Purely extradural spinal meningioma, especially of the "En plaque" variety, usually simulate malignant disease (metastatic diseases and lymphoma) and may result in inadequate therapy, however, the presence of nerve root attachment is even rarer.
Our case report is that of an entirely cervicothoracic extradural en-plaque meningioma (WHO grade 1) with a nerve root attachment (right C7) and intrathoracic extension. We highlighted the issues in diagnosis, operative intervention and long-term follow-up.

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INTRODUCTION

Meningiomas account for about a quarter of all intraspinal tumours and literature has shown that it is the second commonest primary intraspinal tumour [1]. The majority of spinal meningiomas are entirely intradural and this is seen in about 10% of cases.

There is an extradural spinal extension, but attachment to spinal nerve root and/or intrathoracic extension are not a frequent finding [2]. Meningiomas located purely in the extradural space are very rare and, therefore, are not often listed in the differential diagnosis of spinal extradural lesions [2].

We present a case report of purely extradural meningioma "en plaque" of the spine that was noticed to be attached to right C7 nerve root which create a bias for preoperative diagnosis of spinal schwannoma (intradural extramedullary) with extradural extension.

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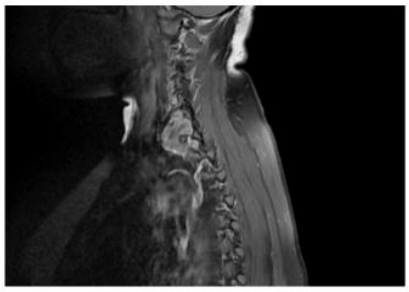
CASE REPORT

We report a case of a 28-year old woman with no previous significant medical history presented with 2-month history of progressive spastic tetraparesis (with muscle power in most muscle groups being 4) associates with right brachio-cervical hyperesthesia involving C6/C7 distribution. Cervico-thoracic magnetic resonance imaging (MRI) done was reported as intradural extramedullary tumour (schwannoma) extending from C4-T1 vertebral level by the radiologist with attachment to right C7 nerve root. Lesion was iso-hypointense on TIWI and enhances on T2WI as shown in IMAGE 1 and IMAGE 2

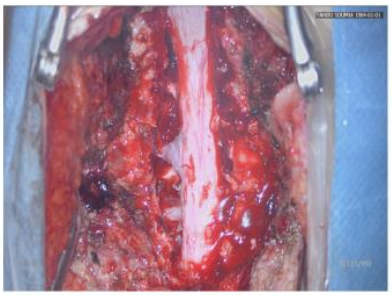
IMAGES 1



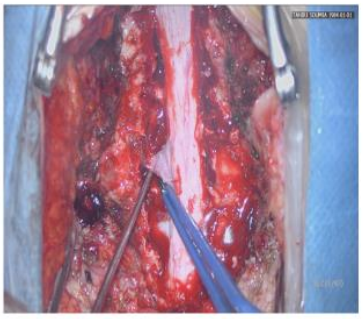
IMAGES 2

perioperatively, the tumour was accessed via C4 –T1 laminectomy, and it was entirely extradural extending from C4-T1 with right C7 nerve root attachment and extending to the intrathoracic region anteriorly as shown in intra-operative images 1 and 2. The tumour was firm, fibrous, highly vascular and dissectible, tumour resected with coagulation of the attachment to right C7 nerve root but the intrathoracic component left in-situ.

INTRAOPERATIVE IMAGES 1

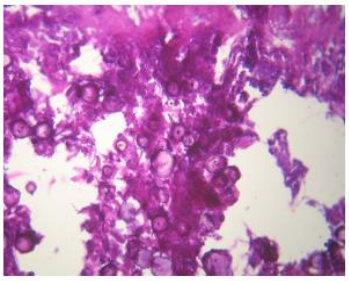


INTRAOPERATIVE IMAGES 2

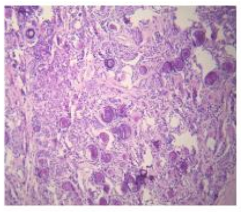


Both frozen section and definitive histopathological/immunohistochemical analyses confirmed mengioma “en plaque” (WHO grade 1) as shown in histology slides below.

HISTOLOGY 1



HISTOLOGY 2



Post operatively, there was an initial loss of sensory modality of right C7 nerve root distribution which improved subsequently in conjunction with motor functions (preoperative tetraplegia) after six (6) weeks of function education and rehabilitation. Follow up is been done regularly using both clinical and radiological tools and there has not been change in improvement achieved postoperatively and also the size of the intrathoracic component.

Discussion

Meningioma is one of the commonest central nervous system (CNS) tumours which is essentially a benign tumour. Majority are intracranial, with about 90% supratentorial, but spinal meningioma account for between 1.2%-12,5% of all meningiomas [3,4,5}. Meningioma arises from arachnoid cap ceil (meningiothelial cell in the arachnoid villi just like any meningioma elsewhere).

The prevalence of extradural meningioma in literature ranges from 3.3% and 21.4% of all cases of spinal meningioma [6].

Tumours occur more in female to male of ratio 4:1 with a peak incidence between 5thand 6th decades of life. Almost about 80% occur in the thoracic spinal region [6]. Majority of these lesions are intradural extramedullary and are placed ventral or ventrolaterally to the dura, However, about 10% do extend to the extradural space [2]. The presence of a "sole" extradural spine meningioma is entirely rare [2]. Common tumours located in the spinal extradural space are metastatic spinal tumour and lymphoma, hence the dilemma in preoperative diagnosis which was also emphasis by Saryedekar et al in the two cases reported by them [7].

Our patient reported above had purely extradural spinal "en plaque" meningioma with right C7 nerve root attachment and intrathoracic extension. The occurrence of "en plaque" variety of meningioma is rarer. Moreso, the presence of nerve root attachment with or without intrathoracic extension is even much more a rarity. Only 16 cases in literature have shown extradural spinal meningioma adhering/proximity to spinal nerve root.

Tuli et al in their case report of a 42-year old lady with T4-T6 extradural spinal meningioma revealed attachment to left T5 spinal nerve root. The presence of extradural meningioma with or without nerve root attachment remain a puzzle and been described as a paradox in view of absence of arachnoid cap cell in the extradural space.

Several theories have been ascribed to the occurrence of an entirely extradural spinal meningioma and possibility of nerve root adherence. Reasons adduced for these in literatures include migration of arachnoid tissue into the extradural space, or extradural spinal meningioma could arise from ectopic or separated arachnoid tissue around the periradicular nerve root sleeve, which is the point of contact of the spinal leptomeninges directly into the dura. This probably explains the attachment to nerve root [2].

Another reason alluded to the occurrence of spinal extradural meningioma is that the periradicular dura which is said to be less thick, may have vestigial remnants of the superficial layer of the embryonal arachnoid mater and villi [6]

Also, it has been suggested that islands of arachnoid tissue that may have migrated into the extradural space can be the source of the meningioma [2,6].

An entirely spinal extradural meningioma especially of the 'en plaque; variety poses a dilemma in pre-operative diagnosis [2, 7]. In this index case, a pre-operative diagnosis of C4-T1 schwannoma of the spine in view of its location and C7 nerve root attachment.

Intraoperative frozen section histopathological analysis is a necessity in view of other possibility such as metastatic spine tumour and lymphoma [8]. In this index case, the frozen section was done which showed a meningioma en plaque. This was done for decision making as regards extend of tumour resection.

The extradural tumour with its attachment to the right C7 nerve root was resected and point of C7 nerve root adhesion was coagulated. However, the thoracic extension was left in-situ. Saryedekar et al in their work shows the importance of intraoperative frozen section with near total resection of spinal extradural meningioma en plaque [7].

This strategy will give the best postoperative outcome, as prognosis depends on the extent of tumour resection [8],

Another consideration in the operation is to decide whether to open the dura or not. However, in consideration of the pathogenesis of the lesion from the dura nerve root sleeve and not from the external part of the spinal dura. It may be necessary to only

peeled off the tumour from the dura. Saryadekar et al in the two cases reported shows that the tumour was stripped off the dura with no durotomy done [7]. Though there is essentially no consensus on whether the dura should be excised or not, However, it is necessary that the dura be opened to rule out intradural extension of the meningioma which is seen in literature to account for about 10% of cases [2].

Tuli et al. reported 12 cases out of 47 supposedly pure extradural spinal meningioma who had durotomy, three (3) of these 12 patients was noticed to have intradural extension. This buttresses the importance of durotomy in hitherto purely extradural spinal meningioma. We had to open the dura to be very sure that there was not intradural component of the tumour as done in standard procedure [6].

Issues relating to long term outcome still remain an item of debate. Issues been debated such as gross total resection versus incomplete resection, benign versus malignant meningioma remain an important factor in considering long-term outcome following operation for extradural spinal meningioma.

Literature has revealed that this tumour may have a local malignant potential despite been a relatively benign tumour. Also, worse prognosis is seen with incompletely tumour resection due to bony involvement or paraspinal extension [2, 3, 8]. In the index case been reviewed, all except the thoracic extension was excised. Tumour has remained static after a regular follow-up period of over 2 years.

Conclusion

Spinal extradural meningioma is a rare tumour, even more with the 'en plaque' variety with spinal nerve root attachment. In view of the prevalence of between 3.3% to 21.4%, it is essential that cases of extradural spinal meningioma should be entertained in conjunction with common tumours such as metastatic spine disease and lymphoma and in doubtful cases Intraoperative frozen section and durotomy are essential in operative decision making either to do gross total resection or partial. For the incompletely resected tumours, long-term follow-up period using both clinical and radiological monitoring tools is essential.

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