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Cord Compression Secondary to Enterobius Vermicularis Infestation

Abstract: An eight-year-old boy presented with a history of severe neck pain radiating to the right shoulder for one week. A well circumferential intra-dural extra-medullary mass arising from the dura on the right antero-lateral side of the spinal cord was seen on magnetic resonance imaging. There was a high intensity dural tail above and below the mass associated and eccentric thickening of adjacent dura of persisting low signal in different images. Surgical exploration revealed a firm pinkish gray, hypovascular mass abutted the dura extending into the right C4 foramina. The dura was thickened not only at the origin of the mass but also a few segment above then and below the lesion. Pathological examination of the tumor revealed enterobius vermicularis inflammatory mass. This report is to our knowledge the first intradural-extramedullary presentation of oxyuriasis. Possible hematogenous mode of involvement of the CSF may further be supported by the present case.

Keywords: Enterobius vermicularis, Cord Compression, Infestation.

Introduction

Parasitic involvement of the spinal cord is very rare. Most such cases have been reported in cysticercosis, schistosomiasis, trypanosomiasis, angiostrongyliasis, and echinococcosis. To the best of our knowledge, leptomeningeal involvement due to enterobius vermicularis has not been reported previously.

Enterobius vermicularis is the cause of the second most common human parasitic infection affecting nearly one tenth of the world population. Humans constitute practically the only host. The infection is found in all age groups and is more prevalent in the cool and temperate zones particularly where people tend to bathe less often and change their underclothes less frequently.^{1,2}

The eggs of enterobius vermicularis are carried through fecal-oral route and very rarely via inhaled dust. The female worm measures 8-13 mm in length and has a pointed tail. The male is much smaller, and measures 2-5 by 0.1-0.2 mm, and has a curved endtail.^{1,2}

Eggs hatch within the body of the host, thus giving rise to a type of infection called retroinfection (or autoinfection). Most of the parasite's development takes place in the large intestine. Worms return to the lumen of the gut and move posteriorly as they mature. Retroinfection may occur when eggs become infective in the perianal area and the hatched larvae reenter the anus.^{1,2} The most frequently reported abnormal migration of F-vermicularis is that along the female reproductive passages. Granulomas induced by the parasite have been reported in genitourinary tract and prostate^{3,4}, peritoneum^{5,6}, perianal area^{5,7}, liver⁸⁻¹⁰ lung^{5,11,12} and CSF.¹³ Enterobius vermicularis may cause peritonitis, abdominal wall abscess¹⁷ and perianal abscess, and can be mistaken for malignancy.

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Case Report

An 8-year-old school boy presented with a history of severe neck pain radiating to the right shoulder for one week. The pain had been unresponsive to usual painkillers including non-steroidal anti-inflammatory drugs. The patient also complained of perianal itching. No other significant history of relevance was obtained.

Physical examination revealed right-sided weakness and a sensory deficit up to the level of the C8 dermatome on the left. Deep tendon reflexes were normal in the upper limbs but they were increased in the lower limbs. Plantar reflexes were equivocal and sphincter tone was intact.

On surgical exploration the spinal cord showed a well circumscribed mass arising from the leptomeninges on the right antero-lateral side of the spinal cord (see below for details). The mass was excised following a C3-C5 laminectomy. On opening the dura, a firm, pinkish grey, hypovascular mass was encountered on the antero-lateral aspect of the cord which abutted the dura and resembled a meningioma. A part of the mass was extending into the right C4 foramina which was probably the cause of the right shoulder pain. In order to extirpate the mass, the dentate ligaments had to be sectioned at the C3/C4 and C4/C5 levels. The dura was thickened not only at the origin of the mass but also a few segments above and below the lesion. The base of the mass was cauterized and the wound was sutured. The post-operative course was uneventful and the patient was discharged from the hospital three days after operation.



Figure 1. A and B. Sagittal postcontrast T1-weighted images show a homogeneously enhancing mass in the mid-cervical region. The lesion is attached to the dura with a broad base and compresses the spinal cord. Associated enhancing dural thickening is also noted.

MRI Findings

Multiplanar T1 and T2 weighted MR images obtained through the cervical and upper thoracic spine in axial and sagittal views, demonstrated a well-circumscribed mass arising from the dura on the right antero-lateral side of the spinal cord. The mass appeared to be attached to the dura by a broad base and caused compression of the cord at C4/C5 levels. However, the visualized parts of the spinal cord showed normal signal intensity and anatomical configuration. There was a high intensity dural tail above and below the mass associated with elongated and eccentric thickening of adjacent dura with persistent low signal in different images. Minimal widening of subarachnoid space above and below the mass and outward displacement of the epidural fat was also noted. The lesion showed homogeneous strong enhancement after DTPA-gadolinium infusion on T1 weighted images. The posterior region as well as paraspinal musculature were normal. Considering these findings which are characteristic of extramedullary intradural mass and the patient's age, our differential diagnosis included meningioma, neurofibroma, solitary metastatic lesion and less likely leptomeningeal infection or inflammation.

Pathologic findings

Histopathological section of the excised tissue showed a predominantly reactive fibroblastic proliferation containing many inflammatory cells mainly of neutrophils, macrophages with abundant eosinophils. There were focal histiocytic aggregates and microgranuloma formation without necrosis. Sections showed what appeared to be pinworms with round and oval structures. The worms harbored a thick external cuticle, a central interstitial lumina and spinous projections characteristic of pinworms. Dense proliferative fibroblastic stromal reaction was seen surrounding the worm contours. There was no evidence of neoplastic changes either in section with or those devoid of worm structures.

Discussion

Although children are more affected by oxyuriasis than adults (20-30% vs 16%)⁴ complicated cases have been mostly reported in the latter. Ectopic worm/egg (granuloma, abscess) have been observed at various sites. The accepted hypothesis for the involvement of the peritoneum and liver is believed to be through the damaged bowel mucosa in both sexes or genital tract in women.¹⁴ Symmers has disputed penetration of the worm through the normal mucosa¹⁴, whereas Faust and DeRuiter have reported mucosal

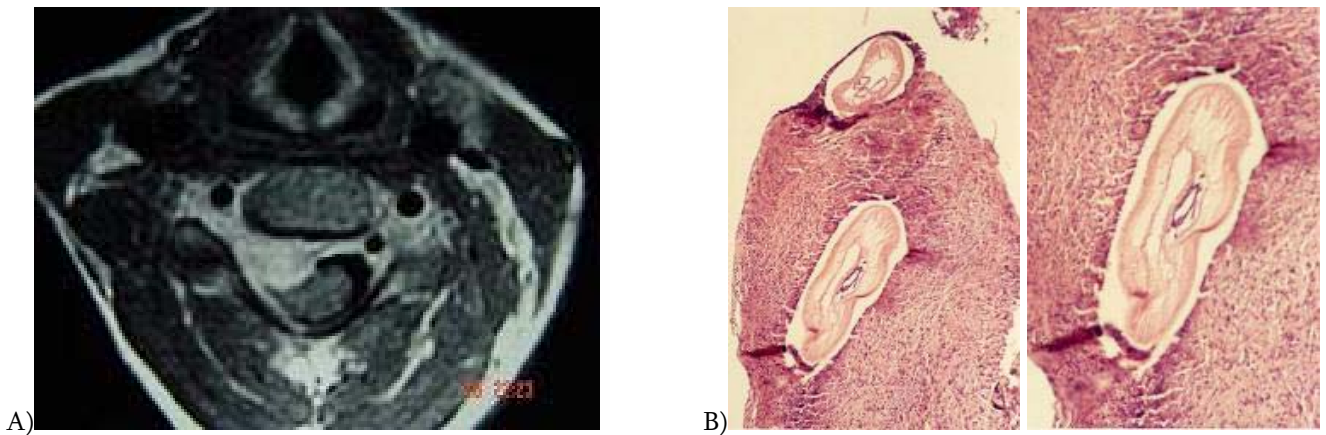


Figure 2. A. Axial post contrast T1 weighted image shows an enhancing mass on the right anterolateral aspect of the cord extending to the right lateral recess. Enhancing dural thickening and displacement of the cord is also seen. B. Section of the tumor shows a few rounded and oval shaped structures indicating cut sections of worm which locally were present. The worm harbored a thick external cuticle and central interstitial lumina and spinous projection, consistent with features found in pinworm.

attachment and partial penetration of the worm in the bowel wall.^{15,16} Susman described a male worm in the peritoneal cavity of a woman with normal bowel mucosa which was later postulated by Symmers to have been due to surgical spillage in that case.⁶ Chandrasoma has reported a male worm in the peritoneal cavity of a woman and 11 male worms within a colonic mass in a man with normal bowel mucosa.⁷ Since the male worms normally die after mating, ascending migration via the genital tract is therefore, unlikely. Chandrasoma while not ruling out the possibility of healed mucosal breakage, has suggested full thickness penetration of the worm in his cases.⁷

Spinal cord diseases are often due to trauma or subdural extramedullary tumors (SDEM) including malignancies, neurofibroma, meningioma, granulomas or tuberculomas.¹⁸⁻²⁰ Infectious causes albeit very uncommon are often due to bacterial abscesses and parasitic infestation.

This case is an unusual presentation of oxyuris involving the cervical leptomeninges. On the clinical context, the young age of the patient would make the diagnosis of meningioma less likely. On MR images, there was a broad base attachment of a well defined mass causing cord compression. In addition low signal on T1 and slightly high signal on T2 weighted images are consistent with intradural-extramedullary lesion. However, strong homogeneous enhancement after DTPA gadolinium infusion on T1 weighted images, helps to exclude other possibilities such as meningocle, enterogenic cyst, lipomyelomeningocele, Tarlov cyst, dorsal dermal sinus and arachnoid cyst. Furthermore, presence of a dural tail and elongated dural thickening lowers the possibility of neurofibroma and schwannoma. These tumors may also be slightly hypointense on T2

weighted images because of fibrous tissue proliferation and cystic spaces. Intratumoral hemorrhage which is more common in schwannoma than neurofibroma can also cause high signal changes on T2 weighted images.¹⁹

Similar to the feature seen in meningioma, our observed mass had broad base attachment, dural tail, well circumscribed mass, hypointense on the T1 weighted and slightly hypersignal on T2 weighted images and considerable enhancement. However associated eccentric elongated dural thickening could be considered as a differentiating feature. In addition, eccentric thickening of the dura at the base of lesion would be an unusual feature for conditions such as pachymeningitis, carcinoma or lymphoma, metastasis and mucopolysaccharidosis deposition.²¹ These conditions would cause concentric narrowing of the subarachnoid space which our case did not have.

Leptomeningeal infection or inflammation would be the other possible radiological diagnosis that could present as a single enhancing subarachnoid lesion or enhancement along the pial margin of the spinal cord. However they usually show low to intermediate signal on T1W and intermediate to high signal on T2 WI. Leptomeningeal infections are best seen on post-contrast imaging. Inflammation and/or infection of leptomeninges can result from pyogenic fungal or parasitic diseases, tuberculosis, blastomycosis, coccidioidomycosis and aspergillosis. In our case we had a single enhancing subarachnoid lesion in association with enhancement along the thickened pial margin which is an unusual radiological association.

Carcinomatous leptomeningeal metastasis presenting as a single enhancing lesion and/or focal or diffuse abnormal subarachnoid enhancement along the pial surface of spinal cord would be another possible radiological diagnosis. However, they usually have

low to intermediate signal on T1W and intermediate to high signal on T2W images. Such lesions commonly show enhancement after injection of contrast. The most frequent subarachnoid metastasis arise from lung carcinoma, breast carcinoma melanoma, lymphoma and leukemia.²⁰ Although we did think of this diagnosis but absence of any other evidence of the malignancy and age of the patient make such diagnosis very unlikely.

Other very rare possibilities which might be considered are Langerhans cell histiocytosis, leptomeningeal hemangioblastomatosis, primary malignant astrocytoma, spinal xantogranuloma and plasma cell granuloma.²⁰ Clearly one could not arrive at a firm diagnosis based on imaging alone without tissue diagnosis.

Lack of bowel disintegrity and absence of the worm or the eggs in the nose, ears, mouth¹⁴, has led same authors to assume yet another route of involvement. Presence of two layers of elastica fibers around the granuloma in Sinniah case, suggested that the worm could have been lodged in the lumen of a vessel.⁵ All the above findings are apparently clues for penetration of the worm through the normal bowel mucosa and subsequent hematogenous spread. Intact overlying skin with no attachment to the mass, speaks against a local penetration. One might conclude that, the worm has probably reached the subarachnoid space hematogenously and lodged in the systemic capillary network. Thus, hematogenous mode of involvement of the liver and lung in previous reports would be further supported by the present case.

This case report describing extramedullary intradural cord compression because of oxyuris would be particularly important in endemic areas and among children. The fact that the disease is medically treatable should highlight the importance of early diagnosis and treatment. High degree of clinical suspicion is of paramount importance for the diagnosis.

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