Retroperitoneal myospherulosis extending to gluteal region through ileum: Presenting as a case of sciatica.

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Myospherulosis is a rare and recently discovered disease apparently caused by the placement of mineral oil, petrolatum-based products or haemostatic agent into different tissues. It can mimic various pathological conditions in different tissues of the body. A sixty year old man presented with left sciatica and a gluteal mass. Imaging showed a retroperitoneal mass extending in gluteal region through the left ileum. Preoperative Fine Needle Aspiration Cytology (FNAC) reported as schwannoma. Patient was managed surgically. Histopathology confirmed myospherulosis. To the author's knowledge, this is the first report of myospherulosis that involved retroperitoneal space and gluteal region through the ileum. In this case presentation clinical features, investigations and surgical management will be discussed.

Keywords: retroperitoneal, gluteal, myospherulosis

Case report:

A sixty year old man presented with diffuse, dull aching pain in left lower abdomen for 3 years and left gluteal region for 2 years. There was severe radiating pain in whole left lower limb more during walking, that made him unable to walk for 1 year. On examination there was a left gluteal mass of about 15x20cm, ill defined margins, hard in consistency, tender, seemed to be fixed to the underlying bone and overlying muscle. Cough impulse was negative but very painful to the mass and to the whole left lower limb. Bulk and power of all groups of muscle in left lower limb were normal. His sensory function and deep tendon reflexes were normal but left hip joint movement was restricted and very painful. Left sided straight leg rising test was positive (100). In the left iliac fossa there was a tender, hard, ill defined, intra abdominal mass which did not move with respiration and in knee- elbow position it became impalpable. Patient could not walk but could stand with much difficulty. Preoperative FNAC from gluteal mass reported as schwannoma. Plain X-ray showed a wide gap in left iliac bone. MRI revealed left sided retroperitoneal tumour extending to the left gluteal region through the ileum (Figure-1A&B). Patient underwent operation under general anesthesia. An S-shaped incision starting from tip of 12th rib curving over the middle of iliac crest extended over the gluteal region and ended just above the greater trochanter (Figure-1C). With the help of iliac osteotomy the dumble shaped tumour (passing through the ileum) was removed. The retroperitoneal portion was well capsulated (Figure-1D) but gluteal portion invaded into the mass irregularly, inferiorly it was on close proximity of the left acetabulum but did not break the articular cartilage. The sciatic nerve was moderately compressed by the mass and there was also some adhesion with the nerve which was released under the
There was no attachment with any other nerves. Gap in the ileum was strengthened with a prolene mesh. The wound closed in layers after leaving a retroperitoneal drain. Histopathology of operative specimen reported as myospherulosis (Figure-2).

Retrogradely, patient had no history of intramuscular injection and other medication in the last thirty years. Three months after the operation, pain disappeared and patient could walk without support but there was some limping on left side. At six month after operation patient could walk normally. Post operative MRI six months after operation showed no residual or recurrent tumor (Figure-3).

**Discussion:**

Myospherulosis was first described in 1969 Africa by McClatchie et al. The name was used based on the suspicion that this might be a curious new fungal infection. It is now known that these structures represent altered erythrocytes. In the first case described in Kenya, the myospherules were round bags of spherules surrounded by a thin, somewhat refractile membrane under light microscopy. The spherules were said to be slightly larger than red blood cells and closely & irregularly packed together. No obvious internal structure was seen, but in some there were small granules or irregular blebs of eosinophilic material. They were located within cysts surrounded by fibrous tissue, histiocytes, and giant cells. Some of the myospherules were ingested by giant cells in...
the cyst wall.³

Myospherulosis can occur in various sites in the body such as muscle, bone, kidneys, liver, retroperitoneum, brain.³ In our case it has involved retroperitoneum, ileum and gluteal region.

The cases reported so far in the literature can be divided into 2 categories: those iatrogenic cases where causality with petroleum-based ointment and gauze packs could be found,³ such as those seen in the paranasal cavity,⁴ ocular cavity,⁵ intracranially,⁶ and in dermal tissue, and second categories are those cases where erythrocytes are altered by endogenous fat or lipids.³ Examples include cases found in subcutaneous tissue, breast tissue, perirenal adipose tissue,⁷ steatocystoma,² and an adjacent benign cystic teratoma.³

Myospherulosis presentation can very according to the location. In the brain it may present like that of a brain tumour.³ In our case patient had presented with local pain, mass and sciatica which is classical presentation of sacral plexus and sciatic nerve tumor. Preoperative FNAC reported schwannoma and radio-imaging also suggested a dumble shaped nerve tumour. Even though histopathology surprised us, per operatively we found only fibrotic attachment with the sciatic nerve in lower gluteal region. In this case, pain might be from pressure over sacral plexus and/or attachment & pressure on sciatic nerve. In such a case, surgical excision is all that needed.

Conclusion:

Though myospherulosis is rare, it can occur at any where in the body without any iatrogenic event, can manifest like space occupying lesion in that part of the body and surgical excision can bring cure.

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