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CASE STUDY

Expect the unexpected–a rare case report of lumbar spinal eumycetoma

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Mycetoma, a neglected disease found in tropical and subtropical countries, is a chronic granulomatous inflammatory disease affecting the subcutaneous tissue. The most common site of infection is the limbs, affecting around 80% of patients. Cases of cranial and spinal mycetomas are very rare. We are presenting an interesting case report of an intradural mycetoma in the lumbar region in an immunocompetent male with no discharging sinus. Notably, a 34-year-old man, presented with symptoms and signs suggestive of lumbar canal stenosis. On imaging with MRI (magnetic resonance imaging), an intradural lesion was observed at the L4-5 level of the lumbosacral spine. Given his history and imaging, an infective etiology, most probably tubercular, was suspected. He underwent L4-L5 laminectomy. During the procedure, pus was discovered extradurally, which was aspirated and analyzed. Upon opening the dura, grayish-black, moderately vascular, suckable grape-like black granules were observed. Sub-total excision of the lesion was performed, and a sample was sent for histopathology. The direct KOH smear showed septate fungal filaments. LPCB (lactophenol cotton blue) preparation revealed hyaline septate filaments with single-celled conidia and a truncated base, which was identified to be *Pseudallescheria boydii*. HPE (histopathological examination) report also concurred that the lesion was an eumycetoma. It is important to note that not all tumors in the lumbar canal are myxopapillary ependymomas, neurofibromas, metastatic bony lesions, or tuberculosis. There is a need to increase awareness and understanding of mycetoma, which would significantly improve disease management, particularly in the endemic areas.

Keywords: eumycetoma, lumbar, intradural, fungal, mycetoma

1. Introduction

Mycetoma, a neglected disease found in tropical and subtropical countries, is a chronic granulomatous inflammatory disease affecting the subcutaneous tissue (1). It can be fungal (eumycotic mycetoma) or bacterial (actinomycotic mycetoma) (2). There are more than 20 species described as causative organisms (3). The most common site of infection is the limbs, affecting around 80% of patients (4). They enter the skin by trivial trauma and present with a pathognomonic triad of multiple sinuses, purulent or sero-purulent discharge containing grains, and a painless subcutaneous mass (4). It is prevalent among field workers such as farmers, individuals of any age, those with immunocompromised status, and in low socioeconomic groups.

Cases of cranial and spinal mycetomas are very rare. Although endemic to India, there are less than 10 reported cases of cranial mycetoma and one case report of spinal mycetoma (5). We are presenting an interesting case report of an intradural mycetoma in the lumbar region in an immunocompetent male with no discharging sinus.

Case report

Notably, a 34–year-old man, presented with low back pain since 4 years, radiating to bilateral lower limbs postero-laterally to ankles since 3 months, claudication distance of only 10 min since 2 weeks, and difficulty clearing ground while walking for 2 weeks. He had no bladder or bowel disturbances. He worked in a local shop and has no history of working in any fields or farm, no history of any trauma in the past, no history of any spinal procedures, no chronic illness, and not immunocompromised. His general and cranial examination were normal; examination of the spino-motor system revealed bilateral ankle dorsiflexion weakness, with sluggish ankle jerks and reduced sensation bilaterally in

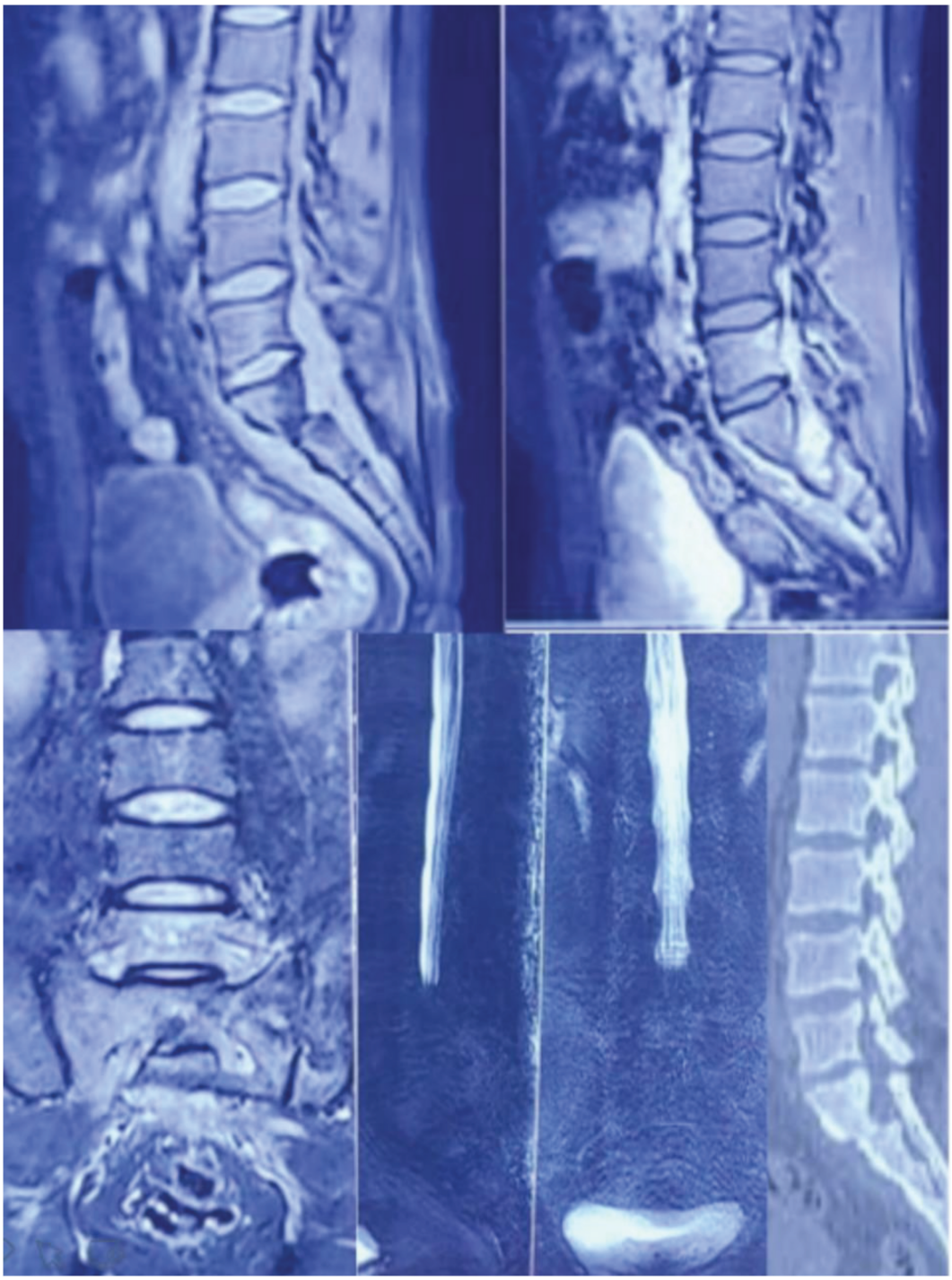

FIGURE 1 | MRI and CT spine imaging showing contrast-enhancing lesion in the lumbar canal causing lytic deformity of the bone, no subcutaneous mass or sinus noted, no root compression.

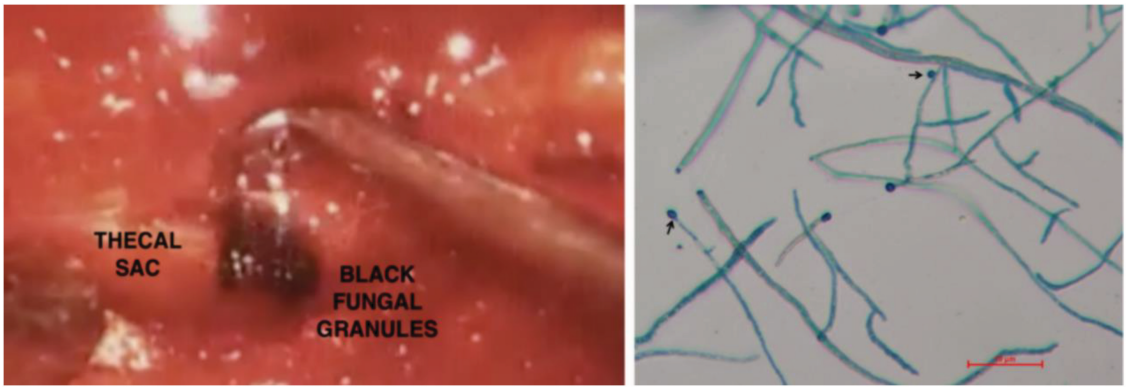

FIGURE 2 | Intraoperative picture showing a black mass in the thecal sac, adjoining smear showing colony of *P. boydii*.

L4-5, S1 dermatomes, more on the left side. Bony swelling with palpitation over the left sacroiliac joint was observed, which was hard in consistency, with no skin changes.

On imaging with MRI (magnetic resonance imaging), an intradural lesion was observed at the L4-5 level of the LS spine causing spinal canal widening at the level and type III arachnoiditis causing bilateral neural foramina narrowing at LS-S1 level. computed tomography (CT) spine showed diffuse sclerosis of the sacrum and left iliac bone with few lytic areas (**Figure 1**).

Given his history and imaging, an infective etiology, most probably tubercular, was suspected. He underwent L4-L5 laminectomy. During the procedure, pus was discovered extradurally, which was aspirated and analyzed. Upon opening the dura, grayish-black, moderately vascular, suckable grape-like black granules were observed. Sub-total excision of the lesion was performed, and a sample sent for histopathology (**Figure 2**). Dura was grossly thickened, so a portion was sent for biopsy.

The direct KOH smear showed septate fungal filaments. LPCB (lactophenol cotton blue) preparation revealed hyaline septate filaments with single-celled conidia and a truncated base (**Figure 2**), which was identified to be *Pseudallescheria boydii* (P. *boydii*). HPE (histopathological examination) report also concurred that the lesion was an eumycetoma.

Post-operatively, the patient began taking voriconazole 200 mg twice a day for 2 weeks, followed by 200 mg once a week, and is attending regular follow up appointments. He experienced significant improvement in pain, and the bilateral dorsiflexion weakness is also showing improvement. He has resumed regular activities.

Discussion

Mycetoma is a chronic suppurative infection affecting skin, subcutaneous tissue, and bones, and is prevalent in tropical and subtropical regions. Chalmers and Archibald classified them into two groups based on the etiological agent—Eumycetoma (fungal) and Actinomycetoma (bacterial) (6).

Patients typically present with a pathognomonic triad of multiple sinuses, purulent or sero-purulent discharge containing grains, and a painless subcutaneous mass. This condition predominantly affects young male adults (7). Although it most commonly affects the lower limbs, cases in unusual locations such as cranial and spinal locations have been documented in the literature. The penetration of the organism through minor trauma or a forgotten injury is often the point of entry (7), though only 20% of patients have a known history of trauma (8). The infection spreads along the fascial planes and leads to the destruction of underlying bone, resulting in osteolytic lesions and discharging sinuses containing the pathognomonic grains, which aids in the diagnosis. Due to late presentation, morbidity is increased. The absence of the discharging sinuses can complicate or delay the diagnosis.

Diagnostic tools include imaging with CT and MRI, tissue diagnostic techniques such as histopathological and molecular tests, classical grain culture, and sero-diagnostic tools. However, many endemic countries lack one or more of these diagnostic tools. Histopathological examination reveals granulomatous fibrous tissue reactions in bacterial infections or fungal hyphae grains in mycotic etiology (9). These grains can also be visualized with various staining techniques such as PAS (periodic acid Schiff), Grocott–Gomori silver, and the classical H&E (hematoxylin and eosin) staining. Treatment is often unsatisfactory on account of the low cure rate, high recurrence, and amputation rate. Surgical excision of the lesion remains the most definitive modality and is supplemented with antimicrobial (actinomycetoma) or antifungal (eumycetoma). Antimicrobials include amikacin at 15 mg/kg/day in two divided doses for 3 weeks along with trimethoprim–sulfamethoxazole at 8/40 mg/kg/day for 5 weeks in cycles, ranging from 5 to 10 cycles (9). Other combination regimes include streptomycin with either dapsone or trimethoprim–sulfamethoxazole given for an average duration of 18 months (ranging anywhere between 6 months and 4 years) (9). Drugs of choice for fungal etiology

include itraconazole or voriconazole given for a prolonged duration of 1–3 years (9).

Spinal eumycetoma is extremely rare, with only four documented cases reported in the literature to date, one of which is from India (10). What makes our case even rare is the absence of a history of any trauma or procedures in the lower spinal regions, a de novo development of the infection in an immunocompetent male with no discharging sinuses in the surrounding region, and the absence of the classical "dot-in-the-circle" sign on T2-W MRI (11).

Pseudallescheria boydii is a eumycete commonly found as a saprophyte in soil and contaminated water. It primarily causes infections in individuals who have experienced near-drowning incidents in such contaminated waters. However, *P. boydii* infections have also been reported in immunocompromised and rarely in immunocompetent individuals who have not encountered such near-drowning incidents.

Conclusion

This case highlights the need to expect the unexpected with lumbar spinal lesions. Not all tumors in the lumbar canal are myxopapillary ependymomas, neurofibromas, metastatic bony lesions, or tuberculosis. Early diagnosis is difficult before the appearance of sinuses or grains. Common misdiagnoses include tumors, chronic bacterial infections, or tuberculosis in a scenarios involving deeper tissue and the absence of discharging sinuses. Hence, to enhance understanding of the disease, achieve early diagnosis, and provide effective treatment, adequate measures should be undertaken in endemic and non-endemic regions to bridge the knowledge gap surrounding mycetoma.

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