

# A Microbial Old Friend with a New Face: A Rare Case of Pyrexia of Unknown Origin and Leukemoid Reaction

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## Abstract

We present a case of a young male, who presented to us with high-grade fever for more than four weeks, refractory seizures, multiple subcutaneous palpable lumps, and evidence of leukocytosis with predominant left shift on the peripheral smear. The classic “starry-sky” appearance on imaging, generalized muscular uptake on positron emission tomography-computerized tomography scan, and positive serology led to a diagnosis of disseminated cysticercosis. He responded to oral steroids. To the best of our knowledge, disseminated cysticercosis presenting as pyrexia of unknown origin and with a leukemoid reaction has never been reported in literature.

**Keywords:** Cysticercosis, leukemoid reaction, pyrexia of unknown origin, starry-sky

## INTRODUCTION

Human cysticercosis is an infection by the larval stage (cysticercus) of the pork tapeworm-*Taenia solium*. Although classically it is acquired through ingestion of undercooked pork, infection in vegetarian children can happen by consumption of food and water contaminated with eggs, autoinfection by eggs refluxing from intestine into the stomach by reverse peristalsis. Persons infected with adult worm may also self-infect with eggs by fecal-oral route.<sup>[1]</sup> Our patient was a pure vegetarian. Few cases of disseminated cysticercosis have been described.<sup>[2-4]</sup> It usually presents as neurocysticercosis (NCC) with headache, seizures, and dementia.<sup>[2]</sup> Although the presence of fever, arthralgia,<sup>[5]</sup> and leukocytosis<sup>[2]</sup> have been reported, its presentation with pyrexia of unknown origin (PUO) and leukemoid reaction is unusual. Our patient had diffuse uptake in muscles and in central nervous system (CNS) on positron emission tomography (PET) scan. Very few case reports of appearances of cysticercosis on PET scan have been reported, such as uptake in lymph nodes and intestine<sup>[6]</sup> and multi-focal uptake in the brain.<sup>[7,8]</sup>

## CASE REPORT

A 28-year-old young male, right-handed, pure vegetarian, resident of Rajasthan was referred to us in July 2013 with a history

of persistent high-grade fever for more than four weeks. He also gave history of seizures inspite of being on two anti-epileptic medications and intermittent generalized headache. He denied any history of tuberculosis, chronic cough, or any weight loss. On examination, he had innumerable just-palpable small subcutaneous lumps all over the body, better felt over the neck, thighs, and paraspinal region without any sternal tenderness, lymphadenopathy, or organomegaly. Neurological examination revealed impaired memory and judgement with exaggerated deep tendon reflexes in the lower limbs and right-sided cerebellar signs without any focal neurodeficit.

His hemogram revealed leukocytosis between 50,000 and 60,000, with predominant left shift without any immature cells and thrombocytosis [Figure 1]. His leucocyte alkaline phosphatase score was high favouring a leukemoid reaction. In view of multiple sterile cultures without any obvious focus, his PET-computerized tomography (CT) scan was planned to determine the most active metabolic focus. Surprisingly, it revealed diffuse uptake in subcutaneous tissues and multiple

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muscles of the musculoskeletal system [Figure 2] and in CNS [Figure 3]. Contrast-enhanced CT of the brain revealed “starry-sky” appearance [Figure 4]. Serum immunoglobulin G cysticercal antibodies enzyme-linked immunosorbent assay was positive (>1 in 800 dilution).

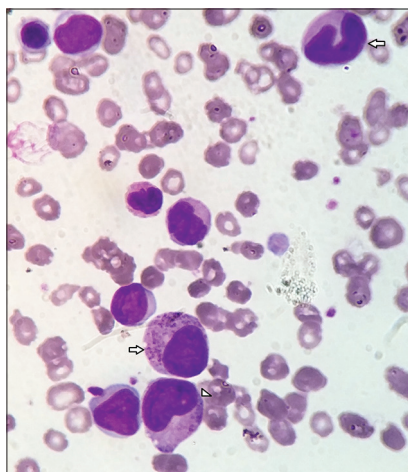
He was given oral steroids (prednisolone 60 mg daily) with antiepileptics (valproate sodium 1g daily, clobazam 5mg twice daily, levetiracetam 500mg twice daily). Albendazole was deferred in view of extensive lesions in the brain and cysticercal encephalitis. He responded dramatically to steroids with resolution of fever, neurological symptoms, and normalization of leukocyte counts within 48 hours. At 2 years of follow-up, he is stable without any new lesions.

## DISCUSSION

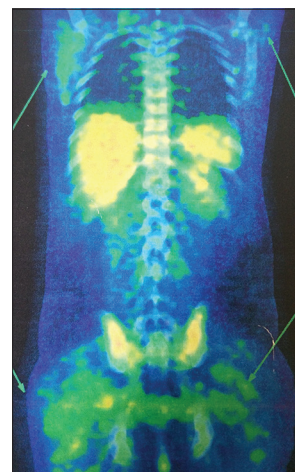
NCC is the most common parasitic disease of the CNS and is linked to accidental ingestion of pork tapeworm (*T. solium*) eggs. It causes about 50,000 deaths per year and is the leading cause of epilepsy worldwide.<sup>[2]</sup>

Common neurologic symptoms include late-onset epilepsy, headache, focal signs, and dementia. Cysticerci can also invade the spinal cord, eyes, subcutaneous and muscular tissues of the body. Muscular pseudohypertrophy is seen more frequently in patients from Asia who have cysticercosis; it accounts for 0.6% of cases in China. This phenomenon is extremely rare in America.<sup>[2]</sup> Our case presented primarily with PUO, seizures, and innumerable subcutaneous palpable lumps.

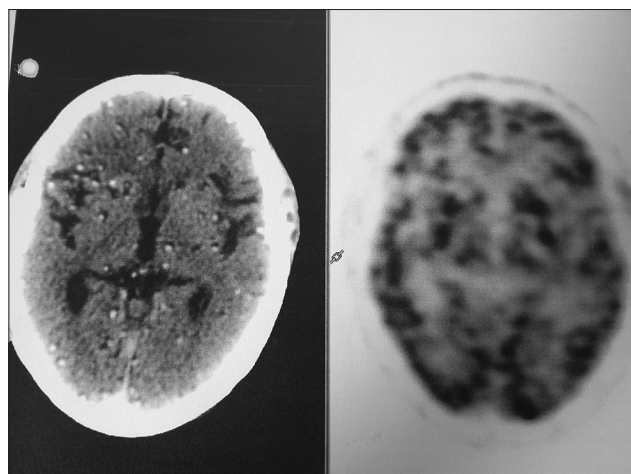
Hematologic abnormalities in patients with NCC include mild leukocytosis and eosinophilia in upto 30% of patients. Stool examination for *T. solium* eggs is positive in only 5%–10% of patients. In a similar proportion of cases, a *T. solium* carrier can be found in the patient’s close environment. Because analysis of the cerebrospinal fluid (CSF) shows abnormalities in 50% or less of patients with NCC, a normal finding on CSF examination does not rule out NCC. Serologic assays, mainly, enzyme-linked immune electron transfer blot, are also commonly used to detect specific antibodies.<sup>[2]</sup> Our case presented primarily with a leukemoid reaction without



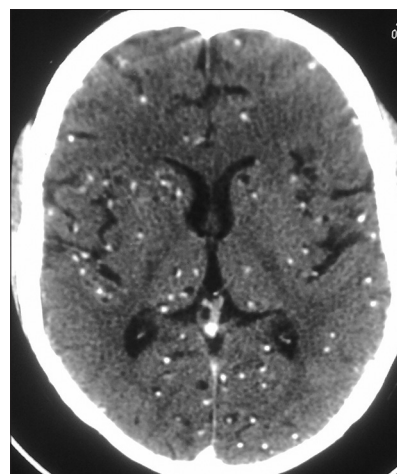
**Figure 1:** Peripheral smear (×100) showing a band form (left pointed arrow), myelocyte (right-pointed arrow) and metamyelocyte (arrowhead)



**Figure 2:** Positron emission tomography-computerized tomography (PET-CT) scan showing diffuse uptake in subcutaneous tissues and multiple muscles of the musculoskeletal system (pointed by green arrows)



**Figure 3:** Positron emission tomography-computerized tomography (PET-CT) of the brain showing diffuse uptake in the brain



**Figure 4:** Contrast enhanced computerized tomogram (PET-CT) of the brain showing “starrysky” appearance

eosinophilia which is unusual. Stool examination for parasites was negative and CSF analysis was normal.

Calcifications in the brain parenchyma are the most common finding in CT scans, and in many cases, the only radiologic evidence of the disease. Hence, CT remains the best screening tool for assessing patients with suspected NCC. Multiple lesions in the parenchyma at different stages are commonly seen in NCC. Small lesions, especially those situated close to bone or within ventricles, may be missed on CT scans. Magnetic resonance imaging (MRI) scan is therefore often added for increased diagnostic sensitivity and accuracy. MRI is also the modality of choice when evaluating patients with intraventricular cysticercosis and when assessing brainstem cysts and small cysts located over the convexity of the cerebral hemispheres. The main disadvantage of MRI is its failure to detect small calcifications. The appearance of cysticerci in brain parenchyma on imaging depends on their stage of development. The cysticercus has four stages of involution: vesicular, colloidal, granular, and calcified. Vesicular cysts appear as rounded lesions with signal properties similar to those of CSF in both T1 and T2-weighted images. The scolex may be seen within the cyst as a high-intensity nodule, giving the lesion a pathognomonic “hole-with-dot” appearance.

When the parasite begins to degenerate, the lesion becomes heterogeneous, and its appearance varies depending on the degree of degeneration. Cysticercotic encephalitis involves multiple cysts in the brain parenchyma associated with severe, diffuse inflammation.<sup>[2]</sup> Our patient had multiple cysticercal granulomas with the classic “starry-sky” appearance on imaging. Very few cases have reported this appearance.<sup>[3,9]</sup> Other differentials presenting with a “starry-sky” appearance on imaging are tuberculomas, chronic lymphocytic inflammation with pontine perivascular enhancement responsive to steroids (CLIPPERS), microabscess, toxoplasmosis, mycotic granulomas, low-grade astrocytoma, and cystic cerebral metastasis. In disseminated NCC, symptoms are related to a space-occupying effect rather than inflammation caused by dying parasites, and in this situation, cysticidal drugs may exacerbate the syndrome of intracranial hypertension.<sup>[3]</sup>

A PET-CT scan was done in our patient for evaluation of PUO. Cases have been described where disseminated cysticercosis has mimicked a lymphoma<sup>[6]</sup> as well as brain metastasis.<sup>[5]</sup> For treatment, albendazole and praziquantel are widely accepted cysticidal drugs. Albendazole is preferred because of its greater availability, higher efficacy, and lower rates of interaction with antiepileptic drugs such as carbamazepine. The standard dose of albendazole is 15mg/kg daily for 10–14 days. Cysticidal drugs damage the parasite and release antigens, triggering an inflammatory reaction that may decompensate the patient. Steroids are therefore used frequently during therapy with anticysticercal drugs in patients with cysticercosis in the brain parenchyma. Surgery is limited to shunt placement for the

control of hydrocephalus in patients with subarachnoid NCC and to the excision of large cysts in the parenchyma in selected patients.<sup>[3]</sup> A meta-analysis found that complete resolution of cystic lesions and reduction of seizure recurrence were significantly better for patients given specific cysticidal therapy compared with patients not given treatment.<sup>[10]</sup> Albendazole or other anticysticercal drugs are contraindicated for spinal and ocular involvement as drug-induced inflammation may result in irreversible damage to the respective organs.<sup>[1]</sup> Although steroids are notorious to cause leukocytosis, our patient’s leukemoid reaction responded to steroids.

## CONCLUSION

Cysticercosis usually presents as NCC with headache and seizures. “Starry-sky” appearance on CT is diagnostic. Steroids with anti-cysticercal drugs are useful in intra-parenchymal NCC.

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## Conflicts of interest

There are no conflicts of interest.

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