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Incidentally Detected Extensive Disseminated Cysticercosis in A Case of Nasopharyngeal Carcinoma

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Abstract: *Taenia solium*, a pork tapeworm, causes a parasitic infection called cysticercosis. It is a frequent source of seizures and neurological morbidity in developing countries. An uncommon presentation of cysticercosis is neurocysticercosis, and till now, less than fifty cases have been noted in the world's literature; most of the cases belong to India. In this study, we have reported an unusual patient of a nasopharyngeal carcinoma presenting with occasional epistaxis and progressively enlarging left submandibular swelling, who has later diagnosed to harbor coexistent disseminated cysticercosis involving multiple sites in the body, namely the brain, extraocular muscles, skeletal muscles, tongue, heart, pleura, peritoneum, aortic wall, seminal vesicles, and scrotal sac. Here we report a 60 years old male patient presenting with occasional nasal bleeding, a headache for 6 months, and swelling in the left submandibular region for 4 months. In the past, the patient had a history of seizure episodes. Nasal endoscopy revealed a mass in the roof of the nasal cavity. A biopsy confirmed the diagnosis of nasopharyngeal carcinoma.Further radiological investigations were made for staging when the patient was detected with extensive rice grain calcifications leading to an incidental diagnosis of disseminated cysticercosis. The first case we know of was detected on an FDG PET/CT (fluorodeoxyglucose-positron emission tomography/computed tomography) scan. The patient was managed with chemoradiotherapy for nasopharyngeal carcinoma and subsequently 400 milligrams of oral albendazole, twice daily, and 20 milligrams of prednisolone, twice daily for three weeks for disseminated cysticercosis. Prednisolone was eventually waned off and terminated. The patient's condition had significantly improved at the 3-month follow-up, and no seizure recurrence was observed. This case report aims to depict the disease's presentation, provide information on this globally prevailing pathogen, and shed some light on the infection's diagnostic workup and therapeutic interventions.

Keywords: Cysticercosis, neurocysticercosis, *taenia solium*, nasopharyngeal, carcinoma

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1. INTRODUCTION

'*Taenia solium*', the pork tapeworm, causes cysticercosis. When tapeworm larvae enter the host, infection occurs. Ingestion of eggs causes them to enter the bloodstream, from where they spread to several systems producing the cysts characterizing cysticercosis. Because cysticercus may be located at any site within the body (most typically in the skeletal muscles and brain), the location and size dictate the clinical manifestation.[1] Campbell and Thomson reported the first instance of cutaneous cysticercosis in India in 1912.[2] Pork tapeworm ova disseminated via the fecal-oral pathway.[3] Cysticercosis constitutes numerous cases of widespread parasite ailments of CNS worldwide, although cysticercosis cutis shows significantly less common occurrence.[4] This disease is prevalent in India, Africa, Mexico, and South America.[5] The mature '*Taenia solium*' adheres to the small intestinal wall and can grow seven meters long in taeniasis. Ova from mature worms in the small bowel of humans are excreted and can survive for weeks.[6] Lesser than fifty cases belonging to disseminated cysticercosis have been noted to date all over the world.[7] Cysticercosis in humans is extremely common in Africa, Europe, and South-East Asia. Cysticercosis is most typically associated with the central nervous system. However, it can also damage the eyes, subcutaneous tissues, liver, skeletal muscle, and, in rare cases, the lung and heart, resulting in various clinical symptoms.[8] Although a few case reports have described disseminated cysticercosis, there are no previous examples involving the heart, lungs, spine, extradural space, and eye lesions within the same patient.[9]

1.1. Disease course

After actively crossing the intestinal mucosa, infective embryos (hatched from ingested eggs) enter the systemic circulation. The liver clears up some of the cysts.[10] Cysts lodge in the capillaries of mostly the muscle and brain tissue, forming immature cysts and then larval cysts, which can take up to three months to reach.[11] The blood-brain barrier protects the cysts from the host's immune response; as previously stated, no inflammatory response is observed as long as the cyst wall, rich in glycoproteins, remains intact.[12] When a parasite dies naturally or due to therapy, an inflammatory response with perilesional edema and calcification occurs.[13]

1.2. Pathophysiological features

Taenia solium advances via one of the following mechanisms.

1. By way of the parasite (because of either obstruction or mass effect)
2. As a result of the inflammatory response (Oedema)
3. By using residual scarring (Formation of granulomas, fibrosis, or calcifications)[14]

Here, we report a case of a 60 years old male patient presenting with occasional nasal bleeding and a headache for six months and swelling in the left submandibular region for 4 months. In the past, the patient had a history of seizure episodes. Nasal endoscopy revealed a mass in the roof of the nasal cavity. Biopsy confirmed the diagnosis of nasopharyngeal carcinoma and was incidentally detected with disseminated cysticercosis while being investigated for metastasis. It is the first case of disseminated cysticercosis detected on an FDG PET/CT (fluorodeoxyglucose-positron emission tomography/computed tomography) scan. The patient was managed with chemoradiotherapy for nasopharyngeal carcinoma. Subsequently, for three weeks, 400 milligrams of oral albendazole, twice daily, and 20 milligrams of prednisolone, twice daily, were administered for disseminated cysticercosis. Prednisolone was eventually waned off and terminated. The patient's condition had significantly improved at the 3-month follow-up, and no seizure recurrence was observed. The aim of reporting this case was to stress that intramuscular cysticercosis should be considered whenever a patient presents with either a nodule or a swelling anywhere on any part of the body in an endemic area. In suspicious cases, ultrasonography could be used as a preliminary examination for ruling out intramuscular 'cysticercosis,' and invasive methods like Fine Needle Aspiration Cytology or biopsy could be used to confirm the diagnosis.[15] A full body FDG PET/CT scan in suspicious cases can be useful when no visible signs exist.

2. CASE PRESENTATION

- **History and clinical examination:** A 60-year-old man presented with a history of occasional nasal bleeding and a headache for 6 months, and swelling in the left submandibular region for 4 months.
- **Medical History:** The patient had a history of seizure episodes for which he had taken phenytoin and phenobarbitone.
- **Family History:** No relevant family history was found.
- **Special tests and investigations:** A diagnostic nasal endoscopy revealed a proliferative mass in the roof of the nasal cavity. A punch biopsy from the mass was suggestive of non-keratinized, poorly differentiated squamous cell carcinoma. (Figure 1)

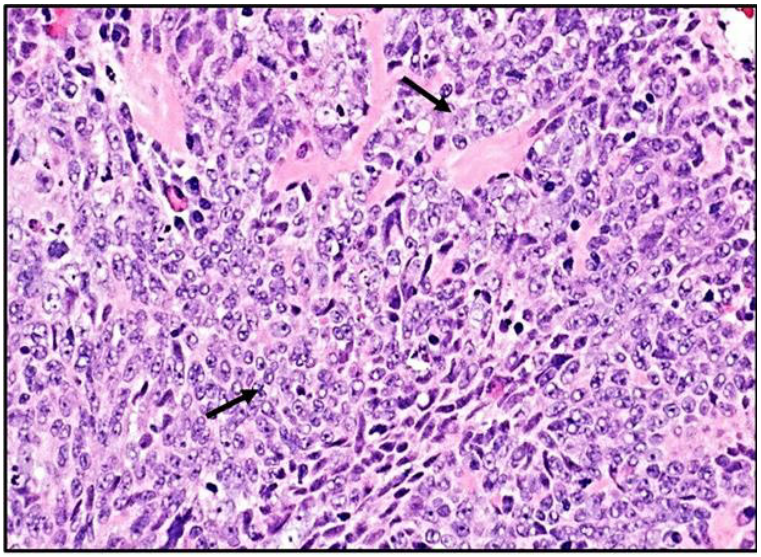

Fig 1: Non-keratinizing poorly differentiated squamous cell carcinoma in high power 40x

Figure 1 shows the histopathological features of a non-keratinizing nasopharyngeal carcinoma depicting a diffuse cellular infiltrate of non-cohesive cells (also referred to as the Schmincke pattern). The tumor cells have moderate eosinophilic to amphophilic cytoplasm, round nuclei, prominent eosinophilic nucleoli, and vesicular chromatin. It shows no significant keratinization. Usually, prominent non-neoplastic lymphoplasmacytic infiltrate accompanies the tumor cells, and necrosis is not commonly found.

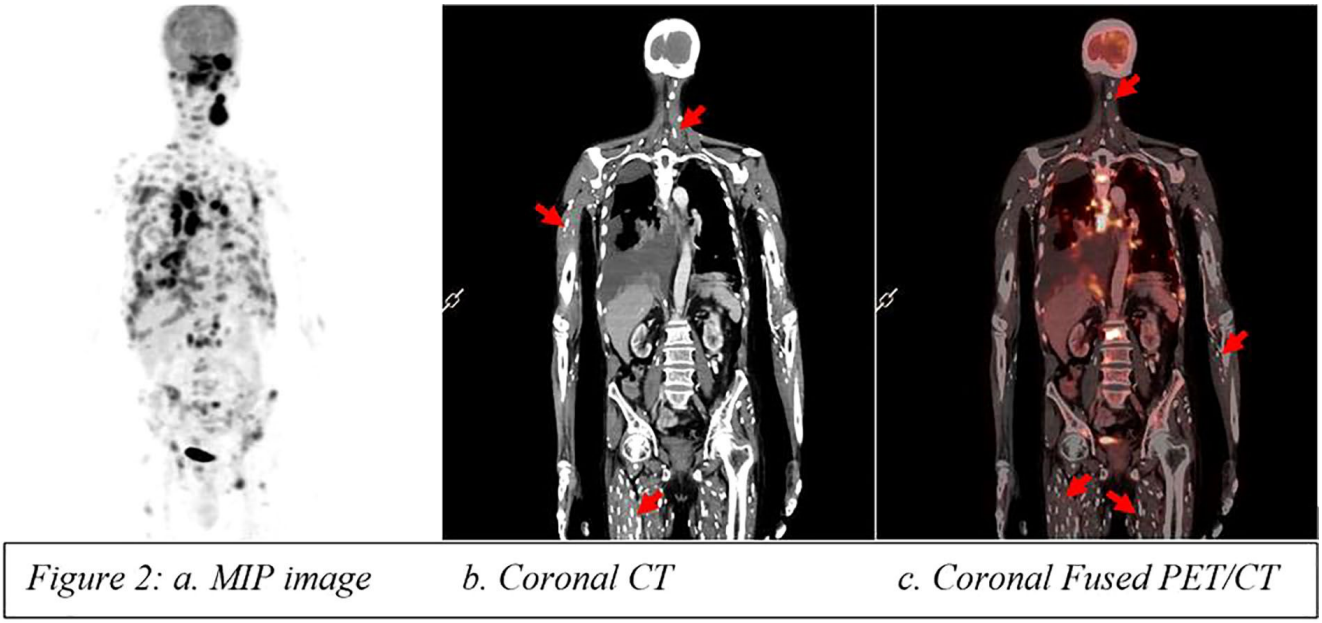

Fig 2: a. Maximum intensity projection image scan b. Coronal computed tomography scan c. Coronal fused positron emission tomography/computed tomography scan

Figure 2 depicts the fluorodeoxyglucose-positron emission tomography/computed tomography scans. *Figure 2 (a) Shows extensively metastatic disease with primary nasopharyngeal soft tissue mass. Figure 2 (b) Shows extensive rice grain calcifications involving the entire visualized skeletal musculature. Figure 2 (c) Shows the fused PET/CT image, which shows no significantly increased FDG uptake in extensive rice grain calcific lesions involving the musculature.* A whole-body FDG PET/CECT scan was done for staging. Maximum intensity projection revealed an extensively metastatic disease with primary nasopharyngeal soft tissue mass. Coronal computed tomography scan showed extensive rice grain calcifications (marked with arrow - Figure 2) involving the entire visualized skeletal musculature. The scan findings suggested a hypermetabolic nasopharyngeal mass with bilateral intraorbital and intracranial extension. Cervical and retroperitoneal adenopathy with extensive skeletal and lung metastases were also noted. Extensive rice grain calcifications involving the entire visualized skeletal musculature and no significant FDG uptake were noted. A uniformly enhancing rounded soft tissue was noted in the left temporal lobe with no FDG uptake, likely representing a nodular granular stage of neurocysticercosis with few other nodular calcified lesions in bilateral parietal lobes of the brain. (Figure 3)

- **Diagnosis:** Histopathological features from the punch biopsy of the nasal mass suggest nasopharyngeal carcinoma (non-keratinizing type). The clinical characteristics and thorough further radiological investigations were done for the staging leading to an incidental diagnosis of disseminated cysticercosis.

- **Management:** After that, chemoradiotherapy was administered to the patient for nasopharyngeal carcinoma and 400 milligrams of oral albendazole twice daily and 20 milligrams of prednisolone twice daily for three weeks. Prednisolone was eventually waned off and terminated.

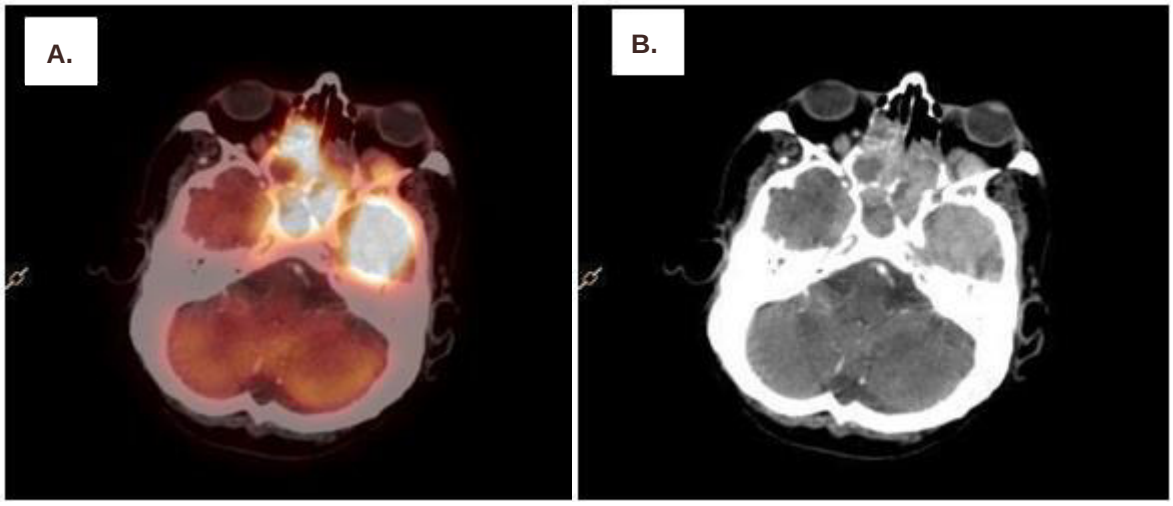

Fig 3:(A, B) shows the intracranial extension of the mass, which is intensely hypermetabolic, as seen on the fused PET/CT images.

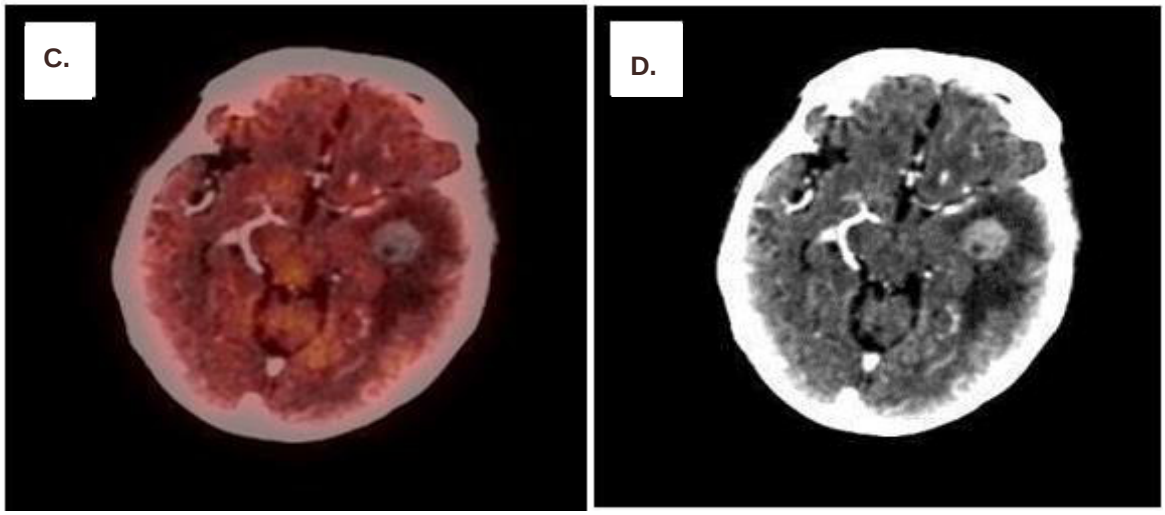

Fig 3: (C, D) Shows uniformly enhancing rounded soft tissue in the left temporal lobe, likely representing a nodular granular stage of neurocysticercosis which shows no significant FDG uptake on the fused PET/CT images.

3. DISCUSSION

Taenia solium's life cycle, parasitology, as well as immunology, are complicated and involve pigs (the intermediate host, which harbors the larval meta-cestode stage), humans (the definitive host, which harbors the adult tapeworm and also acts as unintended intermediate host), and the third one being the ecosystem (which is a source of infection with eggs).[16] Subcutaneous nodules are found in almost half of all cysticercosis patients.[6] But our case had no nodules but was incidentally reported while being investigated for nasopharyngeal carcinoma and was found to have extensively disseminated cysticercosis with granular neurocysticercosis, which is an extremely rare finding. Computed tomography revealed normal brain and muscle appearances. A common myth is that humans can get neurocysticercosis from eating raw pork. Undercooked pork consumption will only result in taeniasis because infected pork contains larval cysts, which progress into adult worms in the human intestine but not the eggs, which end up causing cysticercosis.[17] Cysticercosis in humans results because of disseminated *Taenia solium* embryos to various organs and tissues of the body through the hepatoportal system from the intestine. It most commonly affects subcutaneous tissues, lungs, brain, heart, eyes, liver, and skeletal muscles. Cysticerci can show practically any organ involvement in the body if they have widespread dissemination.[18] Because there is no specific diagnostic evidence on routine blood testing, including peripheral eosinophilia, recognizing neurocysticercosis in the intense setting can be difficult in countries outside of endemic areas. Stool examination has also proved insensitive as there is a long period between exposure to *Taenia solium* eggs and the onset of clinical symptoms.[19] Clinical signs and symptoms, neuroimaging investigations, and epidemiological exposure are used to diagnose.[20] Seizures and headaches are the most common clinical manifestations of neurocysticercosis. Confusion, visual acuity changes, focal neurological signs, stroke, and meningitis are all possible symptoms. Fever is usually absent.[21] Intractable seizures, dementia, muscle hypertrophy, subcutaneous nodules, and a paucity of focal

neurological symptoms or visible rise in intracranial pressure characterize disseminated cysticercosis, particularly until later in the disease's course.[22] The absence of calcification on radiological evaluation in soft tissues and the cranium, as well as the finding of viable cysticercus on either biopsy or autopsy are notable observations, albeit the latter has yet to be seen in sufficient numbers. The most prevalent manifestation of disseminated cysticercosis is muscle pseudohypertrophy, preceded by detectable nodules and seizures.[6] The calcifications represent dead parasites; no further treatment is required; however, they can be used to diagnose disseminated disease. Some authors consider the presence of rice-grain calcifications on plain radiographs to be a minor diagnostic criterion for neurocysticercosis.[23] The presence of rice-grain calcifications may aid in distinguishing neurocysticercosis from its closest comparative, tuberculosis, particularly in endemic areas.[24] Our case showed the presence of numerous rice grain calcifications, which aided in the diagnosis of disseminated cysticercosis. CT scans and MR imaging efficiently detects the cyst anatomical localization and natural history documentation.[25] MR imaging is highly sensitive, even more than computed tomography, in detecting living cysts and scolices in cisternal spaces and ventricles and determining treatment response.[8] Unenhanced computed tomography of muscles can reveal several cysts that stand out against the background of the muscular mass where they are implanted, giving the computed tomography scan the appearance of a 'honeycomb or leopard spots'.[26] Our case presented with neurological symptoms of headache and had a history of seizure episodes and was diagnosed as a case of nasopharyngeal carcinoma, which on further investigations like a full body PET/CECT scan, revealed extensively disseminated cysticercosis with neurocysticercosis. The management of cysticercosis has always been debatable.[5] Albendazole and praziquantel are both effective.[27] Albendazole has more efficacy and is a less expensive treatment for neurocysticercosis than other medications. The standard therapy for systemic, neural, and multiple cysts seems to be either albendazole (15 milligrams per kg every day) given for eight days or more with concurrent steroid administration or praziquantel (50 milligrams per kg every day) given for 15 days.[5]

4. CONCLUSION

To summarise, intramuscular cysticercosis should be considered when a case reports either a nodule or a swelling anywhere on any part of the body in an endemic area. In suspicious cases, ultrasonography could be used as a preliminary examination for ruling out intramuscular 'cysticercosis,' and invasive methods like Fine Needle Aspiration Cytology or biopsy could be used to confirm the diagnosis. A full body FDG PET/CT scan in suspicious cases can be useful when no visible signs exist. A rare complication of cysticercosis is widespread dissemination. Therefore, therapy must be approached methodically.

5. ETHICAL APPROVAL STATEMENT

Written and oral informed consent was taken, and ethical approval was obtained from the institutional ethical committee.

6. AUTHORS CONTRIBUTION STATEMENT

Dr. Ketki Wajpeyi has collected information and prepared the manuscript, which has been thoroughly reviewed and approved by Dr. Mrs. Sunita Vagha. All the authors have read and agreed to the manuscript.

7. CONFLICT OF INTEREST

Conflict of interest declared none.

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