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CASE REPORT Open Access

Cerebral sparganosis: rare parasitic infection of the brain

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Abstract

Spirometra is larval cestode that involve humans as accidental intermediate hosts. Although the incidence of central nervous system infestation with sparganum is low, the diagnosis of the disease can cause delayed with an increased possibility of severe brain damage and neurological deficits. The present case reports a case of a 19-year male student and describes the imaging findings, histopathological characteristics, and management of this rare disease. The patient was treated surgically with good outcome.

Keywords Cerebral sparganosis, Craniotomy, Refractory epilepsy, Spirometra mansoni, Tunnel sign

Introduction

Spirometra is larval cestode that involve humans when a person ingests the plerocercoid which are harbored in frogs, snakes, and chickens [1, 2]. The infection can also spread by drinking contaminated water or contact with the flesh of an infected host to an open wound [3–6]. Humans are accidental intermediate hosts [3, 7, 8], and the disease is most prevalent in southeast and eastern Asia regions [2]. Although the incidence of central nervous system infestation with sparganum is low [9–11], however, as the disease is clinically non-specific, there is chance of delayed or misdiagnosis and an increased possibility of severe brain damage and neurological deficits [2, 4, 12]. The present case reports a case of cerebral sparganosis and reviews the relevant literature.

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Case report

A 19-year male student working as part-time in a restaurant presented with headache on and off, focal seizures involving right upper and lower limbs with secondary generalization and episodes of loss of consciousness since October 2020. He was on tablet phenytoin 100 mg TDS. He had 2–3 episodes of similar seizures 3 weeks ago which was followed by right upper limb weakness. Power in right upper limb gradually improved over 3 weeks to 4/5. There was no history fever, vomiting, cough, or weight loss. His general and systemic examination was unremarkable. Magnetic resonance imaging (MRI) which was performed in 2021 showed a well-defined intensely enhancing lesion in the left precentral frontal cortex measuring 2.5 × 1.6 × 2.1 cm (Previous size was 1.2 × 0.8 × 1.6 cm in year 2020), appearing mildly hypointense on T2 and FLAIR sequence and hypointense on T1WI with subtle blooming on GRE sequence (Fig. 1). There was cystic area measuring about 1.2 × 0.7 × 0.7 cm noted in the anterior aspect of the lesion. The lesion was associated with moderate-to-severe perifocal edema with mass effect in the form of effacement of adjacent sulcal spaces. Blood investigations including HIV, HBsAg, and HCV were negative, and X-ray chest was normal. The patient underwent awake left parietal craniotomy and excision of left posterior frontal lesion. The lesion was surfacing on cortex in precentral gyrus, with flimsy

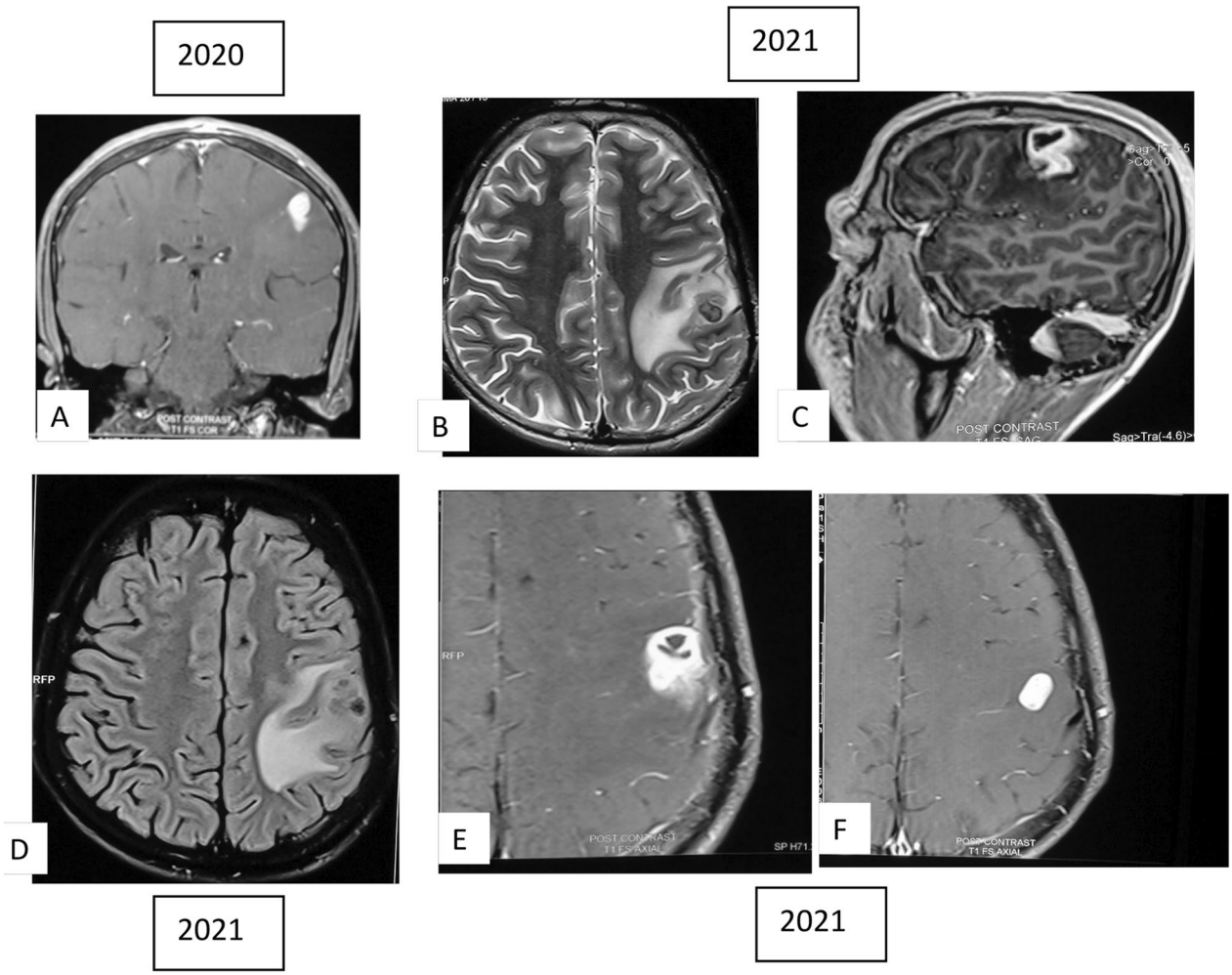

Fig. 1 **A**, **B**, **C** Cranial post contrast MRI and **D** FLAIR; obtained at the onset of headache revealed an irregular enhancement lesion with the perifocal edema in the right frontal lobe with tunnel sign. **E**, **F** Post contrast MRI performed after two days praziquantel treatment showing slight reduction in the lesion and irregular enhancement

adhesions to the dura. Using intraoperative monitoring of right-side motor response and speech response, corticectomy was done around the lesion. The lesion was grayish white firm with nodular morphology, had mild vascularity and there was good differentiation from the surrounding cortex. Gross total excision of the lesion was performed without new neurological deficits. On gross examination, the lesion was gray–yellow, cerebral parenchyma with effaced architecture by the presence of larval forms of spirometra, surrounded by dense diffuse infiltration by polymorphonuclear cells, lymphocytes, plasma cells, eosinophils, and necrosis. There was presence of ill-formed granulomas composed of epithelioid cells and multinucleated giant cells (Fig. 2). Histopathological features were suggestive of cerebral sparganosis involving left posterior frontal cortical region. Post-surgery we gave tab albendazole 400 mg twice daily for 28 days. We have clinical follow-up of patient for 2-year post-surgery. Patient is neurologically stable. He had no seizures after discharge, and anti-epileptic drugs were gradually tapered and stopped.

Optimal treatment for cerebral sparganosis is surgical excision, and treatment with drugs such as praziquantel in dose of 75 mg/kg/day in three divided doses can be considered in case of lesions which are surgically inaccessible. Follow-up MRI imaging is done to see for the reversal of changes, and CSF study for IgG antibodies against *Spirometra Mansoni* was also done.

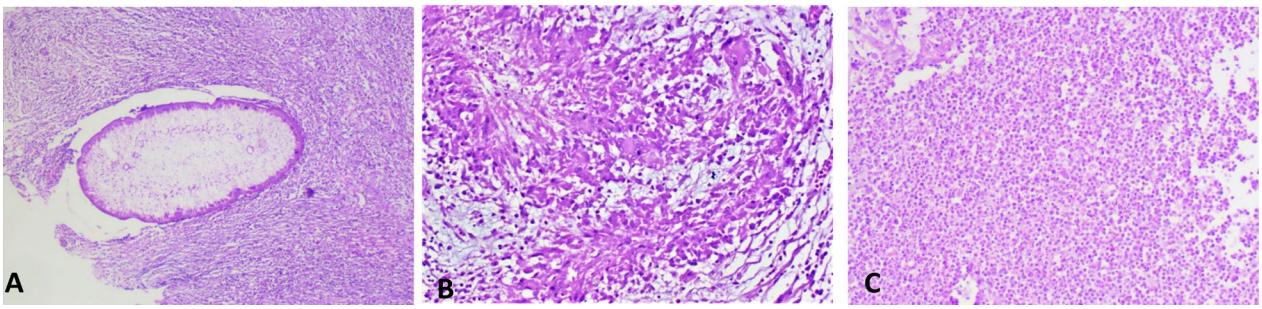

Fig. 2 **A** Larval form of spirometra manson **B** granulomatous inflammation **C** Dense inflammatory infiltrate composed of polymorphonuclear cells, lymphocytes, plasma cells and macrophages

Discussion

Common parasitic infections involving central nervous system include cysticercosis, and less frequent parasitic infections are toxoplasmosis, echinococcosis, and schistosomiasis and rarely paragonimiasis, malaria, toxocariasis, onchocerciasis, Chagas disease, African trypanosomiasis (HAT), angiostrongyliasis, and sparganosis. Human sparganosis although rare but increasingly being recognized as food-borne zoonosis affecting humans who are intermediate hosts. The common sites involved are subcutaneous tissue or muscles; however, uncommonly, the larvae may affect abdominal cavity, pleura, intestines, eyes, brain, and spinal canal. While the brain is involved, clinically the patients may present with seizures, headache, hemiparesis, and sensory disturbances [9, 13, 14]. Acute clinical presentation may mimic subarachnoid hemorrhage or encephalitis [13, 15], and chronic presentation may resemble like granuloma [15]. On CT scan, cerebral sparganosis lesions are usually low-density lesion with unilateral white matter lesions that can change their location, presence of ipsilateral ventricular dilatation, localized cortical atrophy, and nodular or irregular contrast enhancement with spotty calcification [11, 16]. However, spotty calcification may not be present in all the cases [17]. Post-contrast MRI is superior to CT in demonstrating the extent and number of lesions, except the presence of punctate calcifications [1]. The characteristics post-contrast MRI imaging feature of the disease is a "tunnel sign" and the common findings include "bead-shaped enhancement" and the wandering sign [1, 17, 18]. Focal hemorrhages are rare on imaging, though there may be presence of hemorrhage seen on surgery [1].

Cerebral sparganosis can mimic many other lesions involving brain, i.e., chronic cerebral ischemia, brain tumors and other inflammatory granulomas (e.g., mycosis, tuberculosis, or other parasitic infections) [1, 6, 13, 16, 19]. Characteristic imaging appearance of each lesion can help in differentiating among these lesions [1, 16, 19]. Diagnosis can be further confirmed by presence of anti sparganum IgG antibody in the peripheral blood or CSF by ELISA and with molecular diagnostic technique like PCR [17]. Cerebral sparganosis cases can be cured with praziquantel medication alone and has a good prognosis after treatment [18, 20]. However, the management of severe cases and in presence of neurological deficits cerebral sparganosis will need surgical removal of the worm as well as postoperative anti-parasite medication praziquantel or albendazole [2, 18, 20–22]. While performing surgical excision, one need to be careful as the organism may still be alive, and while pulling the worm, it may rupture [9].

Conclusions

Cerebral sparganosis is a rare parasitic infection that involve brain in humans. In both acute and chronic presentations, it can mimic many other CNS pathologies. For accurate diagnosis, high index of suspicion and broad interpretation of clinical and imaging finding are required as early and aggressive intervention will result in good outcome.

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Author contributions
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Declarations

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Competing interests
The authors declare that they have no conflict of interest to this work.

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