CLINICAL REPORT

Ultrasonography Guided Excision of Isolated Cysticercosis of the Temporalis Muscle Causing Intractable Headache: A Rare Case Report

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Abstract Cysticercosis cellulosae is a systemic parasitic infection caused by the larval stage of pork tapeworm, *Taenia solium* which involve humans as either a definitive or secondary hosts. The central nervous system is the most important primary site of involvement. Cases of cysticercosis presenting as an isolated muscle mass is an extremely rare entity and demands documentation. We report an extremely unusual case of isolated cysticercosis of the temporalis muscle causing intractable headache which presented a diagnostic challenge. The condition was surgically treated by ultrasonography guided excision of the cysticercosis swelling in the temporalis muscle. We also emphasize on the role of proper imaging modalities in the diagnosis and treatment of such unique cases.

Keywords Cysticercosis · *T. solium* · Temporalis muscle · Ultrasonography

Introduction

Cysticercosis is a parasitic infection caused by the larval stage of pork tapeworm *T. solium* [1]. This condition is endemic in developing countries of Asian and African subcontinent [2] due to poor sanitation facility and close interaction between humans and animals [3]. The common site of involvement of cysticercosis cysts in humans are central nervous system, eye, striated muscle, subcutaneous tissue, and rarely, in other tissue [1]. Isolated muscular

involvement by cysticercosis is rare [4] and only a handful of cases are recorded in literature. These cases have non-specific manifestations and present diagnostic dilemma to the clinician [5]. We encountered one such perplexing case of isolated cysticercosis of the temporalis muscle which presented with intractable headache since 3 years which was not responding to conservative management of headache. The correct diagnosis was made only after a high index of suspicion and proper radiological investigations and ultrasonography guided excision was performed.

Case Report

A 40 years old female presented to our department with complaints of severe left sided headache since last 2 years which was localized in the left temporal region. The patient had been treated symptomatically for multiple diseases including sinusitis, migraine and temporal arteritis but there was no improvement in symptoms. There was no significant history which was relevant to the condition (Fig. 1).

Examination revealed a barely discernible swelling over the left temporal region which was not mentioned in the previous clinical notes. The examination of ear, nose and throat were normal. Ultrasonography of the suspected site of lesion was performed with a high resolution linear probe using Wipro GE Logic Eg machine which showed a 1.1 cm diameter oval hypoechoic lesion with eccentric small clear fluid in it and surrounding area of fibrosis in the temporalis muscle plain of the area. Colour Doppler supplementation excluded the possibility of its vascular nature. CT scan and MRI of the head revealed ring enhancing cystic lesion with eccentric nodule in the muscle plain of left temporal area characteristic of cysticercosis. Based on the radiological

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Fig. 1 CECT head showing scalp swelling in *left* frontoparietal region of the head having irregular hypodermic lesion with with eccentric enhancing nodule

evidence a diagnosis of cysticercosis of the left temporalis muscle was done. Further evaluation was performed to rule out other foci of cysticercosis in the body. After detailed assessment, no other foci of involvement was found in the patient and the patient was treated as a case of isolated cysticercosis of left temporalis muscle.

The patient was scheduled for excisional biopsy under general anesthesia. The excision was done with the help of ultrasonography guidance in the operation theatre for proper localization and ensuring complete removal of the lesion. The entire lesion was removed in toto and the post-operative period was uneventful. There was no evidence of recurrence or residual disease at 1, 3 and 6 months of follow up (Fig. 2).

Discussion

Cysticercosis is a systemic parasitic infection caused by the larval stage of pork tapeworm *T. solium* [1]. Human beings serve either as a definitive or an intermediate host of the adult tapeworm [6]. Consumption of inadequately cooked pork is the primary cause as pigs serve as normal intermediate host but it can also result from ingestion of tapeworm eggs through contaminated food, water or dirty hands. Cysticercosis is more prevalent in developing countries (mainly India, Indonesia, China, Peru, Africa and

Fig. 2 Flair MRI image of head showing homogenous high intensity oval cystic lesion with tiny eccentric dot in the *left* frontoparietal area of the scalp

Mexico) but may occur in non-endemic areas also [7]. Cysticerci are spherical, milky white cysts containing fluid rich in glycoproteins and a single invaginated scolex with hooklets [8] (Fig. 3).

The most commonly affected sites are subcutaneous tissue, brain, orbit and skeletal muscle [8]. Increased blood supply in these tissues as compared with other organs are hypothesized as reason for preferential survival and growth of cysticerci in these sites [9]. Other less common location for the lodgment of cysticercosis larva are heart, liver, lung and peritoneum [10]. Isolated head and neck muscle cysticercosis is an extremely unusual site of involvement and extensive search of literature revealed that isolated temporalis muscle cysticercosis is even rarer [11].

Fig. 3 An irregular outlined oval cystic lesion with eccentric echogenic nidus and surrounding edema in the scalp suggesting cysticercosis

Diagnosis of isolated muscular cysticercosis is a very perplexing problem due to their rarity and nonspecific manifestations and often present as a diagnostic dilemma to the clinician [12]. Cysticercosis may remain asymptomatic for variable period of time or may present with severe local symptoms. Various types of local clinical manifestations such as myalgic, myopathic, nodular or mass like and the rare pseudohypertrophy have been described in the literature [12]. The various differential diagnosis may include lipoma, fibroma, neurofibroma or intermuscular abscess and it is difficult to diagnose this condition solely on a clinical basis [13].

The importance of imaging modalities in diagnosis of isolated muscular cysticercosis could not be over emphasized. Plain radiography has its role in chronic disseminated form of cysticercosis of skeletal muscle in which diffuse scattered calcifications of rice grain size are seen. However it has got no definite role in diagnosis of acute or vesicular form of cysticercosis as in our case [11]. High resolution sonography is considered pathognomic of cysticercosis and a definitive diagnosis can be made with greater confidence [14]. Vijayaraghavan [14] described four different sonographic appearance of muscular cysticercosis which are: (A) Cysticercosis cyst surrounded by inflammatory mass. (B) Irregular cyst with minimal fluid on one side. (C) Eccentric scolex in the cyst with collection in muscle fibers and (D) Calcified cysticercosis. In our case, high resolution ultrasonography revealed a small cystic lesion with eccentric clear fluid and excluded the possibility of a vascular swelling. The vesicular form of cysticercosis has classical ultrasound appearance of a small cystic lesion with an eccentric nodule and surrounding mild edema. The eccentric nodule represents the scolex of the worm and features are diagnostic for cysticercosis (Fig. 4).

Fig. 4 Excised specimen of temporalis muscle cysticercosis

CT scan and MRI [15] help in showing the location, number and relationship of the cyst to the surrounding structures. CT scan and MRI reveal characteristic appearance of cysticercosis which in our case was a ring enhancing cystic lesion with eccentric nodule. However, Sekhar and Honavar [16] suggested that CT and USG are equally effective in identifying the cyst and the scolex. Arora et al. [11] argued that high resolution sonography provides all information available with MRI and more with regards to muscle pathology.

Treatment of cysticercosis should be based on the manifestations and the anatomical area involved. Medical management is specifically recommended where surgical treatment is risky or not possible as in neurocysticercosis [11]. Praziquantel and albendazole are the two recommended antihelminthic drugs for the treatment of cysticercosis [8]. Surgical excision is recommended for ventricular, ocular, spinal and symptomatic subcutaneous cyst [6] as well defined capsule permits complete excision with unlikely chance of recurrence.

In our patient, considering the severity of the headache we promptly opted for surgical management of the disease. We performed a high resolution ultrasound guided surgical excision of the cyst to ensure accurate localization and complete removal of the disease under general anesthesia. The entire cysticercosis lesion was removed and post-operative period was uneventful. There has been no recurrence after 3 months of post-operative follow up.

Conclusion

Isolated muscular cysticercosis should be considered as a differential diagnosis in cases of solitary swelling of muscles causing a diagnostic conundrum. We also highlight the role of suitable radiological investigation as important diagnostic tool in such cases. Health programs to spread awareness about personal and community hygiene can reduce the incidence of cysticercosis and its varied manifestations in the population.

Compliance with Ethical Standards

Conflict of interest None.

Ethical Consent This article does not contain any studies with human participants or animals performed by any of the authors.

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