

Presentation of cysticercosis of the lateral pterygoid muscle as temporomandibular disorder: A diagnostic and therapeutic challenge

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Abstract

Orofacial pain can often be the chief complaint of many systemic disorders. Cysticercosis involving the lateral pterygoids may cause limitation of mouth opening and may mimic clinical symptoms of a temporomandibular disorder. A 37-year-old female presented with 1-month-old complaint of limited mandibular range of motion. She reported a similar episode a year earlier and was diagnosed with a temporomandibular joint disorder by her primary dentist. Comprehensive intra- and extra-oral examinations were performed, which revealed a limitation of mouth opening accompanied by mild limitation of contralateral excursion. A magnetic resonance imaging revealed a ring-enhancing lesion within the left pterygoid muscle suggestive of cysticercosis. The patient was referred to her primary care physician for further treatment and given physical therapy (stretching exercises) to improve mouth opening. One week later, she developed lesions in the arm and trunk. Further ultrasound imaging of the abdomen and the forearms confirmed the diagnosis of cysticercosis. She was treated with albendazole, physiotherapy, joint stabilization appliance, and had eventual complete recovery. This case emphasizes the importance of diagnosis of a systemic condition that may have serious implications, if untreated, and the importance of a comprehensive evaluation, workup, and multidisciplinary management.

Keywords: Albendazole, case report, cysticercosis, lateral pterygoid muscle, magnetic resonance imaging

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INTRODUCTION

Orofacial pain can be a presenting complaint in a wide spectrum of pathologic conditions ranging from odontogenic infections, underlying systemic conditions, to life-threatening situations such as aneurysms and

malignant tumors.^[1] A significant proportion of these patients may initially present to the dentist for care. Identification of the site and source of the pain can be challenging in many instances. Inadequate diagnosis and failure to refer to the appropriate specialists may render

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the treatment ineffective and result in the progression of the condition.

To our knowledge, case of cysticercosis involving lateral pterygoid muscle causing limitation of mouth opening as the first symptom has not been reported. The objective of this case report is to familiarize dentists with the diagnosis and conservative multidisciplinary management of a systemic condition which may mimic a temporomandibular disorder (TMD) and result in morbidity and mortality if untreated. The article highlights the orofacial pain symptoms reported in cysticercosis of the masticatory muscles and focuses on the process of differential diagnosis in such complex cases.

CASE REPORT

A 37-year-old female presented to the Center for Temporomandibular Disorders and Orofacial Pain, with a chief complaint of limitation of mouth opening and pain on the left side of the face of 1-month duration. The onset was sudden. The patient described the pain in the area of the left cheek and in front of the ear, sometimes radiating to the neck. The patient reported that the pain was related to function, such as talking, chewing, and yawning. The patient also reported a history of a similar episode a year earlier and was diagnosed with a TMD at that time. She was offered home care and stretching instructions (physical therapy) and advised to monitor the pain and limitation of opening. Over-the-counter medication (acetaminophen and ibuprofen) was recommended as needed for the relief of pain. For the limitation of opening, she was instructed to use hot and cold compressions bilaterally in the area of the masseter muscle.

The current pain began spontaneously, the week prior to the evaluation. Pain was frequent (4–5 times a day) and intense in nature, with a duration of a few seconds to minutes. The patient described her pain as continuous and aching with spontaneous attacks. The patient's concerns brought her to the orofacial pain clinic for an evaluation.

The current episode of spontaneous pain of 1 month's duration was not associated with a history of trauma. The patient did not report a history of fever, or neurological problems prior to the onset of the current problem. Her medical, family, and psychosocial histories were noncontributory. She is a vegetarian, but reported eating raw salads at restaurants. She reported that her ability to open her mouth was limited to one-finger width (approximately 4–5 mm). Her primary care physician prescribed a muscle relaxant, carisoprodol 350 mg three

times per day for 2 weeks, and advised her to seek an evaluation for a temporomandibular joint (TMJ) disorder. With carisoprodol, her mouth opening improved to a two-finger width (approximately 10 mm).

The patient's general physical examination revealed a well-nourished adult female, alert, awake, and oriented and cooperative. Her body temperature at the time of presentation was 98.7°F and blood pressure was 124/79 mm/Hg. Cranial nerve screening examination was within normal limits. TMJ and musculoskeletal examination was done in accordance with the DC criteria by a calibrated, American board-certified orofacial pain and TMD specialist. Upon examination, her mandibular range of motion was 13 mm of sagittal opening. Right and left lateral excursions were 3 and 5 mm, respectively. There was mild limitation of protrusive movement (3 mm). Muscle palpation revealed trigger points in the right and left masseter, left temporalis, and left sternocleidomastoid muscles (SCMs) which were also tender upon palpation, but did not completely reproduce the patient's pain. On functional manipulation, the patient reported bilateral pain. The pain was sharp and burning on the left side of the face with functional manipulation. A local anesthetic infiltration was applied in the left posterosuperior alveolar nerve region, which resulted in an increase in opening to 22 mm, and the pain had significantly decreased. The patient does not report pain awakening her from sleep and did not recall of any associated autonomic features.

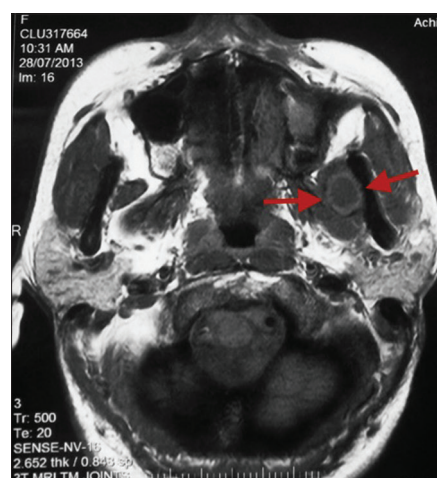


Figure 1: Axial contrast-based T1-weighted magnetic resonance image showing the ring-enhancing lesion suggestive of the helminthic infection, cysticercosis (arrows). The wall of the cysticercus granuloma becomes thick and hypointense and there is marked perilesional edema on T2-weighted images. Cysticercus granuloma shows a ring pattern of enhancement after contrast medium administration. Calcified eccentric scolex is often seen in a cysticercal lesion

A magnetic resonance imaging (MRI) of the brain and TMJ was ordered to rule out intracranial lesions and disc displacement, respectively. The MRI results for the brain were negative. The MRI of the TMJ revealed an unexpected ring-enhancing lesion within the left pterygoid muscle suggestive of cysticercosis [Figure 1]. One week later, she developed lesions on the trunk and arms. The patient was referred to her primary care physician for further tests and treatment. Her primary care physician referred her for an ultrasound of the trunk and arm, which revealed intramuscular cysticercosis in the abdomen and arm [Figure 2]. The patient refused to undergo biopsy. She was managed conservatively using an oral antiparasitic medication, albendazole 800 mg, in divided doses for 30 days. She was followed by her primary care physician and advised to continue stretching exercises advised by the Center for Temporomandibular Disorders and Orofacial Pain for improvement of mouth opening. Stretching exercises comprised of application of moist heat for 3 min followed by gentle stretching using tongue blades until the patient feels a gentle stretching sensation. The tongue blades are kept in position for 1–2 min. The patient was advised to repeat the process three times a day. An intraoral mandibular TMJ joint stabilization appliance was fabricated for the patient. Following appliance calibration, the patient was advised to wear the appliance at night. The patient reported an 80% reduction in pain with the use of the appliance. The patient was then advised physiotherapy for manual release of the trigger points in the temporalis, masseter, and SCMs. One month later, at her follow-up, the mandibular range of motion improved to 35 mm and the patient felt better overall.

DISCUSSION

The complexity of the structures in the head and neck may result in referral of pain to different structures. Often, the source of pain and the site of pain may differ.^[2] Diagnosis of the source of orofacial pain complaints lays the foundation for successful treatment. In certain instances,

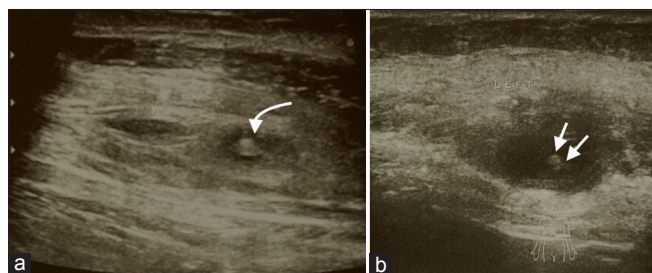


Figure 2: (a) Ultrasound image of the forearm with the curved arrow showing the echogenic scolex and the surrounding inflammation. (b) Ultrasound of the abdomen with straight arrow showing two distinct oval-shaped hypoechoic lesions of the helminthic infection

systemic conditions such as cysticercosis may obscure patient's current complaints or lead to the development of new symptoms in preexisting orofacial pain/TMD patients.

Cysticercosis is a helminthic disease caused by *Cysticercus cellulosae*, the larvae of tapeworm, *Taenia solium*.^[3] Human cysticercosis is a parasitic disease endemic to Asia, Latin America, Eastern Europe, and Africa.^[4] Cases of cysticercosis, which were once limited to endemic countries, are now being reported in various countries around the world due to consumption of infected food, travel, and immigration from endemic areas.^[3]

Pigs are the intermediate host and the infection may spread to humans through ingestion of contaminated food,^[5] water, and internal autoinfection by regurgitation subsequent to reverse peristalsis.^[6] It is disseminated to various organs by penetration of the lining of the alimentary canal, gaining entrance to the systemic circulation.^[6,7]

The condition does not show gender predilection, although there may be variations depending on the endemic areas.^[6] The most common age group is 3–4th decade, although cases have been reported from 1st to 6th decades.^[7,8] The most frequent sites of cysticercosis are subcutaneous layers, muscle in the trunk, upper arm, eyes, neck, tongue, face, and brain.^[7] Oral and maxillofacial region is not a common site in spite of the abundance of muscular tissue and vasculature in the region. Cases in the maxillofacial region including head, neck,^[9] oral cavity,^[8] and masticatory muscles are rare. Among the masticatory muscles, cases of cysticercosis have been reported in the masseter^[10-17] and temporalis muscles.^[18-24] The clinical features of cysticercosis vary depending on the site of infection, number of cysticercosis, and the inflammatory response.^[25] It can be asymptomatic or present with epigastric pain, nausea, diarrhea, swellings, or nodules on the trunk of the body. In approximately 80% of cases, cysticercosis presents as a solitary lesion.^[7,26] Neurocysticercosis is a more serious form and results in neurological manifestations including seizures when the brain is involved.^[27]

Cysticercosis of the temporalis and masseter may be part of disseminated cysticercosis or it may be isolated. The cases of cysticercosis in the temporalis and masseter and their clinical features, diagnostic aids, and treatment are summarized in Table 1. Cysticercosis of the temporalis muscle may present as temporal pain,^[23,24,28] headaches,^[19,23] lump/swelling in the temple,^[23,24,28] and difficulty in masticating food and talking.^[24] Only two cases of cysticercosis of the temporalis presented with swelling, pain, and subsequent limitation of mouth opening.^[18]

Table 1: Cases of cysticercosis in the masseter and temporalis with orofacial pain complaints

Article	Age	Sex	Location	Chief complaint	Other complaints	Duration of symptoms	Diagnostic aids	Diagnosis	Treatment
[1] ²³	43	Female	Left temple	Temporal pain	Headaches	3 weeks	MRI, Serology with ELISA	Temporalis	15 mg/kg/day Albendazole 28 days
[2] ¹¹	22	Female	Right cheek	Recurrent pain	Swelling	3 years	Excisional biopsy, MRI	Masseter	surgical removal and steroids 15 days
[3] ¹⁰	29	Male	Left side of face	Pain	Restriction of mouth opening	3 months	Ultrasonography (USG), MRI	Masseter	Albendazole 15 mg/kg for 28 days and steroids 2 mg/kg gradually tapered over 15 days
[3] ¹⁰	26	Female	Left side of face	Swelling	Restricted mouth opening	20 days	MRI, USG	Masseter	Albendazole 15 mg/kg for 28 days and steroids 2 mg/kg gradually tapered over 15 days
[4] ²⁸	35	Male	Temple	Hard, mildly painful swelling		1 year	CT, FNAC	Temporalis	Albendazole 400 mg/day for 28 days
[5] ¹⁸	16	Male	Right temple	Pain, swelling	Restricted mouth opening	Duration not mentioned	USG, CT	Temporalis	Albendazole 400 mg/twice/day for 14 days and prednisolone tapering
[6] ³¹	21	Female	Bilateral	Progressive headaches		6 months	MRI	Neurocysticercosis with extracranial dissemination involving masseter	Albendazole and steroids and recovery in 2 months
[7] ¹⁶	25	Female	Left	Swelling		3 months	FNAC, USG	Masseter	Albendazole 15 mg/kg body weight/day for 14 days. In addition, an oral dose of methyl prednisolone 2 mg/kg/day was given which was later tapered down over 14 days.
[8] ¹⁹	40	Female	Left	Headaches			USG, CT, US, , excisional biopsy	Temporalis	Excisional biopsy
[9] ²²	35	Male	Right	Restricted mouth opening	Heaviness	1 month	USG, MRI	Temporalis	Albendazole 15 mg/kg body weight/day for 14 days.
[10] ²⁴	50	Female	Right	Pain	Swelling	2 months	USG, MRI	Temporalis	Albendazole 400 mg BD/28 days Crocin for pain
[10] ²⁴	42	Male	Right temple	Swelling	Difficulty chewing food and speaking	Not mentioned	USG, MRI	Temporalis	Albendazole 400 mg BD/28 days Crocin for pain
[11] ¹⁵	12	Male	Right cheek	Swelling		15 days	USG, CT, MRI, histopathological examination	Masseter	Surgical excision
[12] ¹³	18	Female	Right cheek	Swelling	Pain	1 year	USG, MRI, FNAC, excisional biopsy	Masseter	Excisional biopsy

mimicking a temporal space infection.^[22] Cysticercosis involving the masseter may present with pain^[10,11,13] and swelling^[10,11,13,15,16] in the mandible. Only one case of disseminated cysticercosis involving the masseter presented with headaches.^[29] Bhat *et al.*^[10] reported two cases of cysticercosis of the masseter with limitation of opening. However, in one case, it was accompanied with visible facial asymmetry, swelling, and pain and, in the second case, a swelling was palpable on clinical examination. The case reports on cysticercosis of temporalis and masseter muscles unfortunately did not dwell into the characteristics of headache or pain symptoms.

Systemic conditions such as cysticercosis are often underreported and the diagnosis still remains elusive for a majority of health-care providers outside endemic areas owing to lack of awareness and familiarity in managing this

disease.^[3] The diagnosis of cysticercosis may be confirmed with high-resolution ultrasonography, ultrasound-guided fine-needle aspiration cytology, MRI, serum enzyme-linked immunosorbent assay, and histopathology.^[7,22,30] In cases of cysticercosis, complete blood count, erythrocyte sedimentation rate, and liver function tests may be routinely done. Often, they are nonspecific and may not change as in this case. However, in cases where there are deviations, or when the condition is diffuse with difficulty in imaging the entire body, these tests may provide information and may also help predict the recovery status. Additional tests include fecal (stool) examination, also called an ova and parasite test. This test has a limited role since the *T. solium* tapeworm is not viable in the intestine in a majority of the cases, the eggs are intermittently shed, and the morphology of the eggs is similar to the eggs of *Taenia saginata*. Specific tests include enzyme-linked immunoblot transfer blot

assay (which is sensitive for serum in extraparenchymal and multiple lesions), detection of immunoglobulin G antibodies to *T. solium* using Western blot kit (QualiCode), and monoclonal antibody HP10 assay. The results of many of these tests are better when performed on cerebrospinal fluid.^[31]

Cysticercosis is a condition that has a favorable prognosis when managed conservatively. Diagnosis is the cornerstone for effective treatment. Conservative management with anti-helminthic medications has a good prognosis. Albendazole, an anti-helminthic, is commonly used^[24,32,33] at doses of 15 mg/kg in adults. The duration of treatment varies depending on the severity of the disease and may be used for 8, 15, or 30 days. Use of steroids is controversial.^[32,33] Surgical excision of solitary cysts may also be performed if necessary.^[8]

This is probably the first case report of cysticercosis involving the lateral pterygoid muscle resulting in limitation of mouth opening as the first symptom. In addition, this case has certain interesting aspects. The patient is a vegetarian and very few cases of cysticercosis have been reported in vegetarians.^[22,34] We suspect that she acquired the infection through ingestion of either contaminated water or contaminated raw salads. In addition, the limitation of mouth opening was the primary clinical feature at presentation, leading to the assumption that the disease initially affected the lateral pterygoid, causing limitation of opening. In contrast to cysticercosis of the masseter/temporalis which presents with swelling and limitation of mouth opening, mimicking space infections or lipoma, our case presentation initially mimicked a TMD. She presented with limitation of opening, mild limitation of contralateral excursion, with no swelling, thus mimicking disc displacement without reduction in the left TMJ. She later developed swellings in the trunk and arm, indicative of disseminated cysticercosis. However, she did not have any neurological signs or symptoms. Although a specific set of diagnostic criteria for neurocysticercosis have been suggested,^[35] there are no specific criteria for cysticercosis of muscles in head, neck, or for oral cysticercosis. The diagnosis was confirmed by the presence of lesions in the forearm and abdomen suggestive of cysticercosis via positive MRI imaging, ultrasound of abdomen and forearm, and resolution of symptoms after anti-helminthic treatment. Epidemiologically, she is from a cysticercosis-endemic area. Applying the criteria for neurocysticercosis, she had two major, two minor, and one epidemiological criteria suggestive of an absolute diagnosis of cysticercosis.

In the current case, it would have been simple to presume that the patient had been previously diagnosed with TMD and instituted measures for the management of TMD. However, a detailed clinical history and examination revealed that the patient's "familiar pain" was not completely reproduced by muscle palpation. There was only mild limitation in the lateral excursions to the contralateral side. In contrast, in instances of disc displacement without reduction in the TMJ, there is major limitation of contralateral excursive movements.^[36] Further, the local anesthetic block failed to completely eliminate the pain and thus raised warning signs that further investigations were necessary.

In many countries, TMD patients are referred to the prosthodontists for evaluation and treatment. In such instances, it is mandatory for the prosthodontist to perform comprehensive evaluation and arrive at an accurate diagnosis before initiating treatment. Failure to accurately diagnose secondary causes mimicking TMD symptoms can result in the institution of inappropriate treatment, morbidity, and mortality.

Red flares should be noted when there are deviations from normal clinical signs and symptoms in an orofacial pain case. They should be thoroughly investigated and appropriate referrals should be made when appropriate. A simple paradigm for diagnosis of orofacial pain condition is represented in Figure 3.

Although rare, the diagnosis of cysticercosis should be considered in the differential diagnosis of patients living

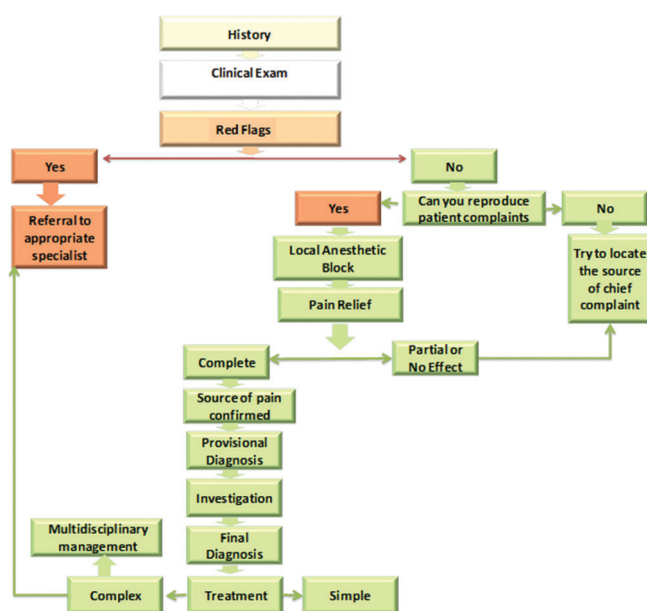


Figure 3: The paradigm for differential diagnosis of complex cases of orofacial pain

in endemic areas and presenting with orofacial pain or symptoms mimicking TMD. Prompt diagnosis and immediate treatment is of paramount importance to avoid morbidity and mortality.

CONCLUSION

A case with a cysticercosis of the lateral pterygoid muscle presenting with a limitation of mouth opening and mimicking the symptoms of left TMJ disc displacement without reduction was described. The importance of a comprehensive evaluation with a detailed case history supplemented with a detailed physical examination and diagnostic testing was described. The responsibility of the dentist to each patient includes a correct diagnosis and an institution of appropriate referral to the physician. Knowledge of the unusual causes of orofacial pain in head and neck is essential. Systemic conditions can obscure the existing orofacial pain complaints or result in sudden changes in their symptoms. This should raise a red flag to the treating dentist and such changes warrant a prompt referral to an orofacial pain specialist for a complete workup. Treatment should be instituted after definite diagnosis and multidisciplinary management may be required in complex cases.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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