Case Report

**Neurotrichinosis. A case report from Turkey**

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**Abstract**

Trichinosis is a parasitic disease which develops after ingestion of undercooked meat contaminated with larvae of Trichinella spiralis. Undercooked pork and its products are the most common source of human infection. Although trichinosis is a worldwide disease it has not been reported from Turkey until 2003. This is probably because of the limited ingestion of pork in this country due to religious reasons. We report a 40 year old woman with tachypnea, tachycardia, diarrhea, fever, myalgia, subconjunctival hemorrhage, periorbital and facial edema with myositis in the masseter and tongue muscles and CNS findings such as delirium, seizures and hemiparesis. who was diagnosed with possible neurotrichinosis.

Key words: Trichinosis, neurotrichinosis, Turkey

**INTRODUCTION**

Trichinosis is a parasitic disease which develops after ingestion of undercooked meat contaminated with larvae of Trichinella spiralis (6). Undercooked pork and its products are the most common source of human infection (12).

Trichinosis is characterized by three phases (11). The enteric phase ranges from 10 days to several weeks. The new larvae migrate to the circulatory system and make their way into all parts of the body but survive only in the striated muscle where they become encysted and eventually calcify (1). The invasive phase lasts from weeks to months and is mainly characterized by eosinophilia, fever, myalgias, muscle weakness and tenderness, subconjunctival hemorrhages, periorbital and facial edema (12). The cardiac, pulmonary and central nervous system involvement may occur during this period. The third stage is the convalescent stage which corresponds to encystment and repair (1).

Blood assay discloses leukocytosis particularly eosinophilia in the acute phase and an increase in the total Ig E level. Muscle enzymes are also elevated (12). A positive test for specific serum antibodies for Trichinella spiralis or a parasitological investigation by muscle biopsy is needed to confirm the diagnosis.
Central nervous system (CNS) involvement has been estimated at about 10-24% of symptomatic cases (12). Mortality is 10-20% in cases with CNS involvement (7). Symptoms such as headache, vertigo, delirium, seizures, aphasia, psychiatric changes as well as focal signs have been reported (8). Several pathogenetic mechanisms have been proposed for cerebral involvement such as mechanical obstruction of arterioles and capillaries by larvae, hypersensitivity or immunological reaction with vasculitis, toxemia from muscle or larval breakdown products, but the exact mechanism remains unknown (6).

CASE REPORT

A 40 year old woman had been in good health until December 2002, when she noticed pain and swelling in her right cheek. A masseter muscle biopsy disclosed accumulation of inflammatory cells (chiefly lymphocytes, few neutrophils, no eosinophils) among the muscle cells and muscle fiber degeneration. She was diagnosed with inflammatory myopathy and was treated with prednisolon 16 mg tid with some clinical response. After a few days of myalgia, diarrhea and fever she developed a confusional state with meaningless speech, inability to recognize her children and hallucinations and was admitted to the hospital on February 20, 2003. She had no localizing signs on her neurological examination and was diagnosed with delirium. The next day she developed severe breathing difficulty, together with a puffy face, a swollen tongue and seizures. She was tachycardic (140/min), tachypneic (30/min) and dyspneic with a fever of 39 C. She also had periorbital and facial edema, bilateral subconjuctival hemorrhage and tongue edema. She was unable to talk due to her swollen tongue and severe dyspnea. Her neurological examination that day revealed a right hemiparesis and bilateral unresponsive plantar responses.

She had a normal CBC with an ESR of 46mm/h. The serum CPK level was 1029 U/L (N<145), and LDH level was 1161 U/L (N<480). There was an increase in total IgE 296.9 IU/L (N 1.5-100). The other biochemical evaluations were normal, including the tests for vasculitis such as C3, C4, RF, ACA Ig G, ACA Ig M, ANA, Anti-ds DNA, C-ANCA, P- ANCA.

An LP was performed on admission with normal pressure, 2 cells/mm3, normal glucose and protein content. A second LP on the 5th day had similar results.

A brain MRI scan demonstrated a small low signal intensity area in T1 weighted and a corresponding high signal intensity area in T2 weighted sequences in the left putamen.

A tongue biopsy revealed severe, chronic inflammation with eosinophil and leukocyte infiltration, degeneration of muscle fibers and mild interstitial fibrosis.

The patient was diagnosed with probable neurotrichinosis and antihelmintic therapy was begun with albendazole 400 mg bid and pulse steroid 1000 mg. A tracheostomy was performed and mechanical ventilation instituted due to respiratory insufficiency caused by her swollen tongue, with possible contribution by laryngeal and/or intercostal muscle involvement.

Her clinical condition improved dramatically as of the second day of therapy. She was successfully weaned from the ventilator and her tracheostomy was closed on February 28. Antihelminthic therapy continued for 15 days, and after 5 days steroid treatment was continued with oral prednisolone 40 mg/day which was slowly tapered after 2 months. She was discharged after 1 month of admission with no sequela.

DISCUSSION

We describe a patient with tachypnea, tachycardia, diarrhea, fever, myalgia, subconjuctival hemorrhage, periorbital and facial edema with myositis in the masseter and tongue muscles and CNS findings such as delirium, seizures and hemiparesis.

A confirmed case of trichinosis is defined as a patient presenting one or more of the followings; fever, myalgia or facial edema with either a Trichinella positive muscle biopsy or a positive serological test (3,6,12). A probable case has been defined as a patient with at least three of the following signs or symptoms: fever, myalgia, facial
edema and eosinophilia > 1,000 cells/mm3 (3).

One of the main criteria, eosinophilia was not found in our patient. Eosinophilia has been reported to occur in 8-76% of cases depending on the stage of the disease. Wang Z.Q. et al reported a normal eosinophil count in 22 of their 29 patients who were treated with steroid before the diagnosis of trichinosis (13). Our patient had been treated with steroid before admission, which might be an explanation for the lack of eosinophilia. Also it has been reported that eosinophil count may be depressed in severe cases.

The most commonly involved muscles in trichinosis are tongue, masseter and pharyngeal, ocular and intercostal muscles, which all were affected in our case (2). Serological tests for trichinosis are not available in Turkey because of the rarity of the disease. Although we found inflammatory myopathy in both right masseter and tongue muscles, we were not able to see a larva in any of the muscle biopsies. According to the criteria defined by Ancelle T. et al our patient is a case of probable trichinosis (3). Another important point was the good clinical response to antihelmintic therapy which occurred in the first 2-3 days of treatment.

Until 2003 there had been no reported case of human trichinosis in Turkey, however it was reported in wild boars (10). The main source of infection, pork, is not a common food in Turkey due to religious reasons. However it is known that beef products may become inadvertently adulterated with pork or other wild animals during processing (9). After the first submission of this paper there has been an outbreak of trichinosis including 542 patients in Izmir Turkey (4). The infection source has been traced to a single seller of "cig kofte", a traditional raw meatball usually made with uncooked veal but which in this case was adulterated with pork (5).

We were not able to track our patient’s source of infection. Regardless of the source of the meat, the key factor in preventing trichinosis is cooking the meat thoroughly. It is very important to recognize this treatable condition which the clinical course can be highly variable, ranging from no apparent infection to a fatal disease.

Nörotrişinozis: Bir olgu sunumu

ÖZET

Trişinozis, Trişinella spiralis larvası ile kontamine olmuş etin yenilmesi sonucu insanlara geçen bir parazit hastalığıdır. En sık kaynak olarak domuz eti gösterilirken farklı etler de bulaşıcılık bildirilmiştir. Santral sinir sistemi tutulumu %10-24 olgunsu görülürken mortalite bu olgularda %10-20 ye kadar ulaşmaktadır. Trişinozis tüm dünyada görülen bir hastalık olmakla birlikte 2003 yılına kadar Türkiye’den insan trişinozis vakası bildirilmemiştir. Bu sebeple 40 yaşındaki diare, taşıkardi, taşıpne, ateş, myalji, subkonjunktival hemoraji, periorbital ve fasil ödem, masseter ve dil kasında miyozit ile birlikte delirium tablosu gelisen, solunum yetmezliği sebebiyle solunum cihazına bağlanan ve antihelmintik tedavi sonucunda 1 ayda sekelsiz iyle 40 yaşındaki bayan hastayı, muhtemel nörotrişinozis olgusu olarak sınımayi uygun gördük.

ANAHTAR SÖZÇÜKLER: Trişinoz, Nörotrişinoz, Türkiye

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