CASE REPORT

INTRAOSSEOUS PROLIFERATIVE SPARGANOSIS PRESENTING AS A PATHOLOGICAL FRACTURE: A CASE REPORT AND REVIEW OF THE LITERATURE

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Abstract. We report a case of 63-year-old male, who presented with pathological fracture of left distal humerus 3 weeks previously. The radiographic findings showed an ill-defined permeative osteolytic lesion of the left distal humerus. Incisional biopsy and debridement was done; pathological examination revealed a folded cestode larva with calcareous corpuscles in the bone and soft tissue, and increased eosinophils. IgG antibody tests for sparganosis were positive. The patient refused to have surgery for internal fixation and placement of an endoprosthesis.

Keywords: proliferative sparganosis, distal humerus, pathological fracture

INTRODUCTION

Sparganosis is a parasitic infection caused by the plerocercoid larvae of diphyllobothroid tapeworms belonging to the genus *Spirometra* (John and Petri, 2006). The typical spargana are white, flat, ribbon-like worms. Their size varies from a few millimeters to several centimeters in length. Sparganosis is transmitted to humans by drinking water contaminated with copepods containing *Spirometra* larvae, by consuming the raw flesh of a second intermediate host, such as frogs or snakes, or by placing a poultice infected with plerocercoid larvae on an open wound (Hughes and Biggs, 2002). Clinical manifestations are diverse. Spargana can invade the eyes, central nervous system (CNS), breast, and subcutaneous tissues, causing diseases such as blindness, paralysis, and even death (Cho *et al*, 1975; Kim and Lee, 2001; Boonyasiri *et al*, 2013, 2014). The majority of cases occur in Southeast Asia and eastern Africa. The largest number of cases occur in Korea and Japan. To the best of our knowledge, only one case of intraosseous proliferative sparganosis arising in the long bone has ever been report from Thailand (Daengsvang and Tansurat, 1947).

We report a case of sparganosis involving the long bone of a man who presented to Srinagarind Hospital with a pathological fracture.

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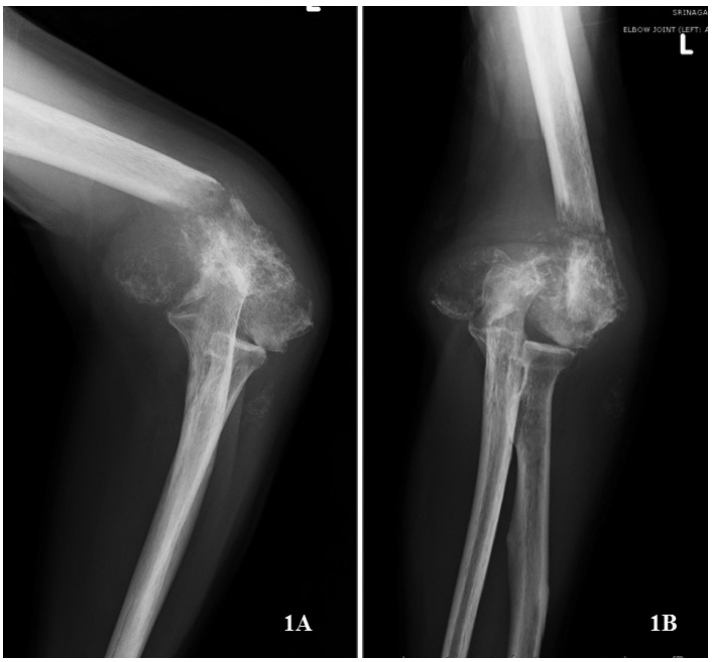

Fig 1–Plain radiographs of the left elbow: anteroposterior view (1A) and lateral view (1B) showing an ill-defined permeative osteolytic lesion involving in the left distal humerus with pathological fracture.

CASE REPORT

A 63-year-old male farmer presented to Srinagarind Hospital with pathological fracture of left humerus 3 weeks previously. Five months prior to admission, he had pain and swelling of his left elbow with no history of trauma. He was diagnosed as having a bone tumor at a community hospital in Mukdahan Province, northeastern Thailand and referred to Mukdahan Provicial Hospital. Although he was treated with an arm slab, tramadol, piroxicam and orphenadrine citrate/paracetamol, he still had limited range of motion of the left elbow but no pain. After that he was lost to follow-up.

He had a history occasionally eating undercooked meat and raw fish but denied ingesting raw snakes, frogs or birds or placing a poultice of raw snakes or frogs on an open wound.

On physical examination, his temperature was 37.1°C, his pulse rate was 70/min, his respiratory rate was 18/min and blood pressure was 128/71 mmHg. The patient was alert, mildly pale and non-icteric. His left elbow was swollen but not warm or erythematous. His elbow was flexed and had limited range of motion due to pain. The other joints were normal.

A complete blood count revealed a hemoglobin of 9.3 g/dl, a white blood cell count of 5,000/mm^3 with 49% neutrophils, 29% lymphocytes, 10% monocytes, 11% eosinophils, 1% basophils and a platelet count of 232,000/mm^3. His erythrocyte sedimentation rate was 46 mm/hour. His serum creatinine level was 1.4 mg/dl and his serum calcium level was 8.5 mg/dl. Stool examination revealed *Strongyloides stercoralis* larvae and *Opisthorchis viverrini* eggs.

Plain radiographs of the left elbow showed an ill-defined permeative osteolytic lesion involving the left distal humerus with a pathological fracture (Fig 1A-1B). A three dimensional computerize tomography (CT) scan showed a pathological fracture of left distal humerus with a permeative osteolytic lesion destroying the entire medial and lateral epicondyle of left humerus with intraarticular extention; no periosteal reaction; there was decreased trabeculation of entire proximal shaft of the humerus (Fig 2).

After admission, an incisional biopsy was performed and systematic debridement was done. Pathological examina-

Fig 2–Three dimensional-computerize tomography (3D-CT) scan showing a pathological fracture of the left distal humerus with a permeative osteolytic lesion destroying the entire medial and lateral epicondyle of the left humerus with intra-articular extension.

tion of the debrided matter revealed a solid section of a folded cestode larva with calcareous corpuscles in the bone and soft tissue, mixed inflammatory cells and increased eosinophils (Fig 3A-3D). Praziquantel 25 mg/kg orally three times a day for 2 days was given to treat the *Opisthochis viverrini* and albendazole 400 mg oral twice a day was given to treat *Strongyloides stercoralis*. IgG antibody tests for cysticercosis using ELISA (Intapan *et al*, 2008) and sparganosis using ELISA and immunochromatography (Yamasaki *et al*, 2014) were done: the result was positive for sparganosis and negative for cysticercosis.

The orthopedist recommended the patient has surgery for internal fixation and an endoprosthesis placed but the patient refused. He was then discharged from hospital with a long arm slab.

DISCUSSION

Human sparganosis is a zoonotic disease which can be classified into two types, non-proliferative sparganosis caused by an infection with canine and feline tapeworm, genus *Spirometra,* and proliferative sparganosis caused by an infection with *Sparganum proliferum* (John and Petri, 2006). There is no clinical difference between the two species except *S. proliferum* is migratory (Miyazaki, 1991). The first case of human proliferative sparganosis was described in Japan (Ijima, 1905). The most common sites this parasitie infects are the skin and subcutaneous tissues (Miyazaki, 1991). Skin involvement is usually multiple or disseminated (Miyazaki, 1991). Intraosseous proliferative sparganosis is an extremely rare parasitic disease. Parasitic invasion of the bone has been found in two cases reported by Liao *et al* (1984) and Nakamura *et al* (1990). The involved bones were the lumbar spine and the iliac bone in which the parasites destroyed the bony cortex and medullary cavity.

The first case of sparganosis reported from Thailand was in 1943 (Daengsvang and Tansural, 1974); from then until 2012 there were 64 reported cases (62 non-proliferative and 2 proliferative) (Anantaphruti *et al*, 2011; Boonyasiri *et al*, 2013, 2014). Nearly half the cases were cutaneous and one-third were ocular. Ocular sparganosis was prevalent until 1990 due to the traditional medical practice of applying frog flesh topically as a poultice to treat sore eyes. The drastic decrease in ocular sparganosis cases indicates a

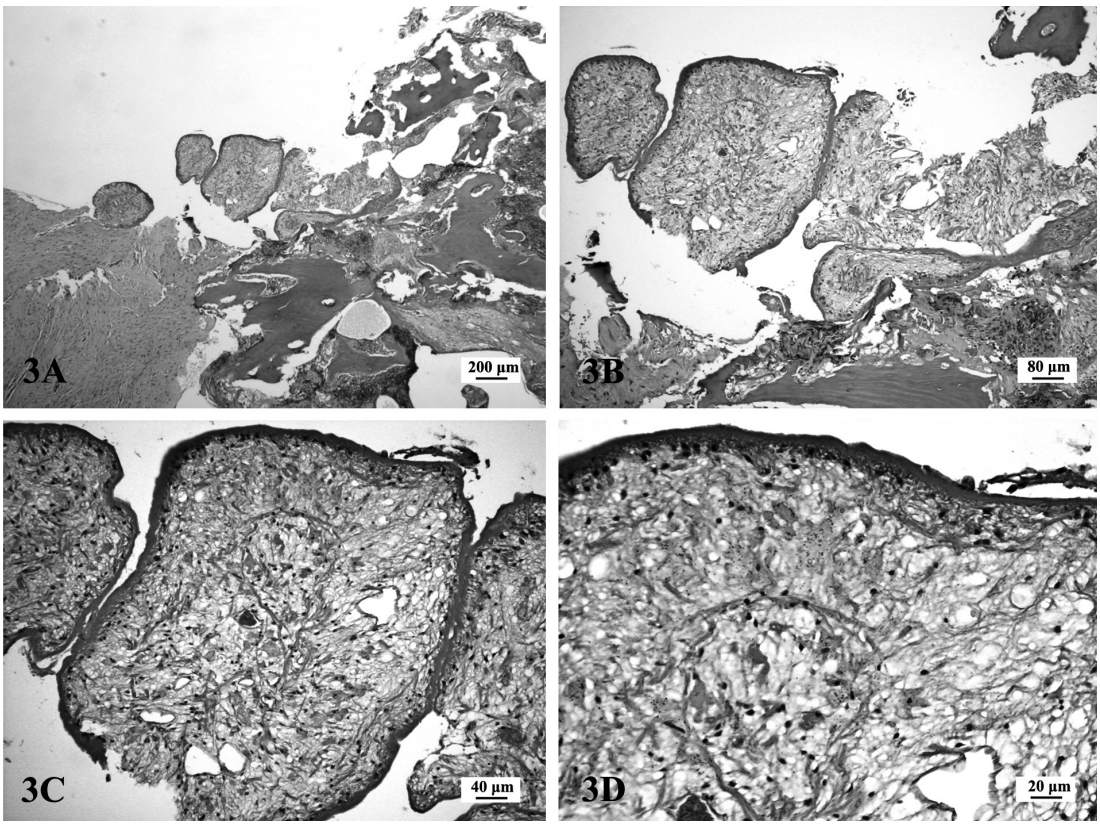

Fig 3–Pathological examination (hematoxylin and eosin stained) revealing a solid section of a folded cestode larva with calcareous corpuscles in the bone and soft tissue, mixed inflammatory cells and increased eosinophils (3A, 40x magnification; 3B 100x magnification; 3C 200x magnification and 3D 400x magnification).

changed in this practice and a decline in the use of traditional medicine. The relative increase in subcutaneous cases, indicates sparganosis remains an important food-borne zoonosis in Thailand. The first case of intraosseous proliferative sparganosis occuring in a long bone was reported by Settakorn *et al* (2002). The patient described here denied ingestion of raw snakes, frogs or birds or placing a poultice of raw snakes or frogs on an open wound but did admit to eating undercooked meat and raw fish.

Sparganosis is typically diagnosed following surgical removal of the worms, although the infection may also be diagnosed by identification of eosinophilia or identification of the parasite in a tissue specimen. If a pathological diagnosis is not feasible, the antisparganum ELISA test may be used to determine exposure (Walker and Zunt, 2005).

A pre-operative diagnosis made by exposure history and painful, migratory, subcutaneous nodules is not common. This patient had eosinophilia and an elevated erythocyte sedimentation rate (ESR) which suggested a parasitic infection but not sparganosis specifically. Our case was diagnosed from post-operative tissue pathology, ELISA and an immunochromatography test.

Treatment of sparganosis is with praziquantel 120 to 150 mg/kg body weight over 2 days, although with limited success (Walker *et al*, 2005). Praziquantel has no effect on adult worms in the central nervous system. Cerebral sparganosis requires surgical excision of the parasite. In general, infestation by one or more sparganum larvae is often best treated by surgical removal. There is no available treatment for proliferative sparganosis. Attempts at surgical removal of *S. proliferum* have been unsuccessful because of widespread dissemination of the larvae (Kim *et al*, 1997; Murata *et al*, 2007).

In summary, we present a case of pathological fracture caused by intraosseous proliferative sparganosis which was misdiagnosed as a bone tumor. This highlights the need to have a strong index of suspicion for this parasitic infection in tropical countries in patients with suspected pathologic fracture.

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