

# Echinococcal Osteomyelitis of the Humerus: A Case Report of an Indigenous Case in Thailand

Jariyasomboon S, MD<sup>1</sup>, Pradniwat K, MD<sup>2</sup>, Treetipsatit J, MD<sup>2</sup>

<sup>1</sup> Anatomical Pathology Unit, Ratchaburi Hospital, Ratchaburi, Thailand

<sup>2</sup> Department of Pathology, Faculty of Medicine Siriraj Hospital, Mahidol University, Bangkok, Thailand

**Background:** Echinococcal osteomyelitis is a rare localization of echinococcosis. Regarding the skeletal site, the spine is most commonly involved. Echinococcal osteomyelitis of the humerus is uncommon, even in endemic areas. In Thailand, only 28 cases of echinococcosis have been reported. Musculoskeletal involvement was identified in two of these cases. None involved the humerus.

**Case Report:** In the present report, the authors describe a rare case of indigenous echinococcal osteomyelitis of the humerus in an 18-year-old Thai woman from Ratchaburi province who presented with pathologic fracture.

**Conclusion:** This particular case does suggest that echinococcal osteomyelitis, despite its rarity, can be encountered in non-endemic areas including Thailand. Since its clinical and radiographic manifestations can mimic tumors and other inflammatory conditions, the disease might be overlooked. Histopathologic examination of the lesion is crucial for making diagnosis of echinococcal osteomyelitis.

**Keywords:** Echinococcal osteomyelitis, Osseous echinococcosis, Osseous hydatidosis, Hydatid disease of bone, Osteomyelitis

**J Med Assoc Thai 2019;102(2):227-30**

**Website:** <http://www.jmatonline.com>

Echinococcosis or hydatid disease can be caused by species of the tapeworm, *Echinococcus*. Human echinococcal infection occurs worldwide and results from ingestion of contaminated water and food or handling live animals that act as definitive hosts, such as dogs. After being ingested, the eggs hatch in the small intestine and release minute hooked embryos that penetrate through the intestinal wall and then are transported by the bloodstream. When the organisms reach their destination, they produce hydatid larval cysts. The infection usually affects liver and lung, whereas bone infection or echinococcal osteomyelitis is rare and occurs in 0.2% to 4% of cases<sup>(1)</sup>.

In Thailand, echinococcosis is a rare disease. Between 1936 and 2017, only 28 cases have been reported in the English literature<sup>(2-5)</sup>. In order of frequency, liver, lung, abdominal cavity, kidney,

spleen, foot, and jaw were involved. Echinococcal osteomyelitis of the long bones has not been previously reported in Thai population. The authors, therefore, reported a rare case of indigenous echinococcal osteomyelitis of the humerus, to address its clinical, radiologic, and histopathologic manifestations.

## Case Report

An 18-year-old Thai woman from Amphoe Chom Bueng, Ratchaburi province, Thailand, presented at the local hospital with sudden pain in her left arm after she had fainted and fallen onto the ground. Her past medical history was unremarkable. On physical examination, the left arm was deformed, swollen, and tender on palpation. Fracture was suspected. Therefore, the patient was referred to Ratchaburi Hospital for further management. At Ratchaburi Hospital, plain film of the left humerus (Figure 1) showed a geographic lytic lesion without sclerotic rim at the midshaft. A displaced pathologic fracture was also noted. No soft tissue calcification was observed.

Based on the clinical and radiographic findings, the clinical impression was a benign bone tumor, such as fibrous dysplasia or aneurysmal bone cyst, with

## Correspondence to:

Treetipsatit J.

Department of Pathology, Faculty of Medicine Siriraj Hospital, Mahidol University, 2 Wang Lang Road, Bangkoknoi, Bangkok 10700, Thailand.

**Phone:** +66-2-4196520, **Fax:** +66-2-4114260

**Email:** [jitsupa.tre@mahidol.ac.th](mailto:jitsupa.tre@mahidol.ac.th)

**How to cite this article:** Jariyasomboon S, Pradniwat K, Treetipsatit J. Echinococcal Osteomyelitis of the Humerus: A Case Report of an Indigenous Case in Thailand. *J Med Assoc Thai* 2019;102:227-30.



**Figure 1.** Radiograph of the left humerus illustrating a geographic lytic lesion without sclerotic rim at the midshaft. A displaced pathologic fracture is also noted. No periosteal reaction is observed.

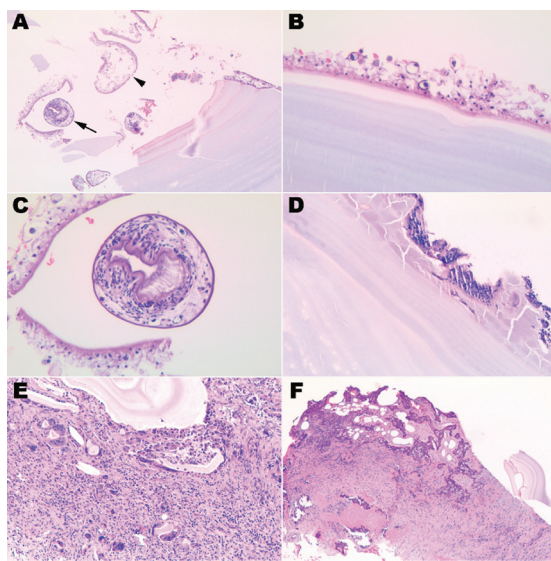
pathologic fracture. Extended curettage of the bone lesion was performed. Thereafter, open reduction internal fixation (ORIF) with plate and screw was applied to the fracture. Tissue from the curettage was submitted for histopathology.

Histologic examination of the submitted material (Figure 2) showed multiple fragments of acellular laminated cyst wall. Some of these fragments were observed in association with thin and eosinophilic nucleated germinal cyst wall and protoscolices. Few fragments of fracture callus and reactive fibrovascular tissue with prominent granulomatous inflammation with foreign body-type multinucleated histiocytes were noted in the vicinity of the parasite. Echinococcal osteomyelitis was diagnosed on the basis of the aforementioned histopathologic findings.

The patient was treated with albendazole. Additional assessment on the patient's risk of infection revealed that she has owned a dog. There was no history of traveling to endemic areas of echinococcal infection. Nine months after surgery, removal of plate and screw was performed when the fracture was healed. No evidence of the disease was documented in the healed fracture site and elsewhere.

## Discussion

Echinococcal osteomyelitis, a rare localization of echinococcosis, is caused by *E. granulosus*. The



**Figure 2.** Histopathology of the curettage specimen. (A) Fragments of *Echinococcus granulosus* comprising acellular laminated cyst wall (right lower corner), thin eosinophilic nucleated germinal cyst wall (arrowhead), and protoscolex (arrow). (B) The acellular laminated cyst wall present in association with nucleated germinal cyst wall. Note calcareous corpuscles within the nucleated germinal cyst wall. (C) Protoscolex liberated from a ruptured cyst. (D) Foreign body-type multinucleated histiocytes surrounding the parasitic cyst wall. (E) Prominent granulomatous inflammation with foreign body-type multinucleated histiocytes and (F) fracture callus in the surrounding host tissue. Hematoxylin & Eosin, original magnifications  $\times 100$  (A, E, and F),  $\times 400$  (B, C, and D)

bone infection occurs by hematogenous seeding. The frequently involved skeletal structures include the spine, pelvis and femur<sup>(6-10)</sup>. The humerus is much less frequently involved in both endemic and non-endemic regions of the world<sup>(6,7,11-17)</sup>. Cases with echinococcal osteomyelitis of the humerus in the English literature is summarized in Table 1. Regarding the non-endemic areas, echinococcal osteomyelitis of the humerus has been reported once in the English literature<sup>(13)</sup>. The present case represents another case of echinococcal osteomyelitis of the humerus that occurred in non-endemic areas, and the first reported case in Thailand.

After being lodged within the bone, *Echinococcus* produce hydatid larval cysts that fill the spaces between the bony trabeculae and eventually result in their resorption of the neighboring cortex. These changes are typically reflected in a radiograph as single or multiple expansile lytic lesions with cortical thinning. Usually the lesion lacks a periosteal reaction.

**Table 1.** A summary of cases with echinococcal osteomyelitis of the humerus in the English literature

Authors	Year	Number of cases	Age	Gender	Nationality	Clinical presentation	Location
Endemic regions*							
Aslan et al. <sup>(12)</sup>	2001	1	35	Female	Egypt	Pain and swelling	N/A
Markonis et al. <sup>(14)</sup>	2001	1	17	Male	Greece	Painless, enlarging mass	Shaft, especially the distal part
Ozkan et al. <sup>(16)</sup>	2008	1	56	Male	Turkey	Pathologic fracture	Metaphysis extending to the diaphysis
Laubscher et al. <sup>(17)</sup>	2008	1	41	Male	South Africa	Pathologic fracture	Proximal diaphyseal region
Nourbakhsh et al. <sup>(15)</sup>	2012	1	N/A	N/A	Iran	Non-union fracture	N/A
Bracanovic et al. <sup>(6)</sup>	1971 to 2010	3	N/A	N/A	Serbia	Pathologic fracture	Metaphysis, extending later to diaphysis
Gun et al. <sup>(7)</sup>	2005 to 2016	1	N/A	N/A	Turkey	N/A	N/A
Non-endemic regions							
Chari <sup>(13)</sup>	2007	1	35	Male	India	Painful swelling	Entire scapula and upper third of the humerus
The present case	2018	1	18	Female	Thailand	Pathologic fracture	Midshaft

N/A=not available

\* Referring to South America, Africa, Europe, Middle East, and Australasia<sup>(18,19)</sup>

Osteosclerosis may be observed in the late stage of the disease<sup>(20)</sup>. In the present case, these characteristic radiographic features were present. However, the exclusive localization of the infection in the diaphysis was unusual for hematogenous osteomyelitis. It is because of the uncommon site of involvement, in conjunction with rarity of the disease in Thailand, that this case was initially misdiagnosed as a benign bone tumor. In this situation, histopathology plays a crucial role in making diagnosis of echinococcal osteomyelitis.

Histopathologic diagnosis of echinococcosis caused by *E. granulosus* can be rendered by identification of the acellular laminated cyst wall, which may be present in association with the thin, eosinophilic, nucleated germinal cyst wall and/or protoscolices. Parts of the parasite can induce reaction in the surrounding host tissue. In echinococcal osteomyelitis, the parasite induces resorption of bony trabeculae and production of reactive fibrous tissue infiltrated by a mixture of inflammatory cells including lymphocytes, eosinophils, and plasma cells. In addition, granulomatous inflammation with foreign body-type multinucleated histiocytes is observed in the vicinity of the parasitic parts<sup>(1,21)</sup>. In this case, the authors made the diagnosis of echinococcal osteomyelitis based on the identification of the parasitic parts in the histologic sections of the curettage specimen. In order of frequency, the acellular

laminated cyst wall, the thin, eosinophilic, nucleated germinal cyst wall, and protoscolices represented the parasitic parts encountered in the histologic sections. Regarding tissue reaction in the vicinity, granulomatous inflammation with foreign body-type multinucleated histiocytes was the most prominent reaction identified. In addition, reactive fibrovascular tissue with lymphocytic, plasmacytic, and neutrophilic infiltrates and fracture callus were focally noted. The findings in this case address histopathologic characteristics of echinococcal osteomyelitis.

## Conclusion

This particular case does suggest that echinococcal osteomyelitis, despite its rarity, can be encountered in non-endemic areas including Thailand. Since its clinical and radiographic manifestations can mimic tumors and other inflammatory conditions, the disease might be overlooked. Histopathologic examination of the lesion is crucial for making diagnosis of echinococcal osteomyelitis.

## What is already known on this topic?

In Thailand, echinococcosis is a rare disease. Less than 30 cases have been reported since 1936. Liver and lung are the two most commonly involved organs. Musculoskeletal involvement was found in less than 10%. None of these reported cases involved long bones.

## What this study adds?

This report described the first indigenous case of echinococcal osteomyelitis of the humerus in Thailand. Due to its rarity, histopathologic examination of the lesion is crucial for making diagnosis of echinococcal osteomyelitis since its clinical and radiographic manifestations can mimic tumors and other inflammatory conditions.

## Conflicts of interest

The authors declare no conflict of interest.

## References

1. Rosenberg AE, Kattapuram SV, Nielsen GP. Infection of bone. In: Kradin RL, editor. Diagnostic pathology of infectious disease. 2nd ed. Philadelphia, PA: Elsevier; 2017. p. 369-403.
2. Morakote N, Thamprasert K, Lojanapiwat B, Muttarak M. Cystic echinococcosis in Thailand with a special note on detection by serology in one family. Southeast Asian J Trop Med Public Health 2007;38:796-8.
3. Riangchan P, Suankratay C, Wilde H, Thanakit V. Hydatid disease of the liver: the first indigenous case in Thailand and review of the literature. J Med Assoc Thai 2004;87:725-9.
4. Waikagul J, Dekumyoy P, Anantaphruti MT. Taeniasis, cysticercosis and echinococcosis in Thailand. Parasitol Int 2006;55 Suppl:S175-80.
5. Warnnissorn N, Uprasertkul M, Atisook K, Sirivatanauksorn Y, Limawongpranee S. Alveolar echinococcosis in a Thai patient after migration to an endemic area in Central Europe. Travel Med Infect Dis 2006;4:34-7.
6. Bracanovic D, Djuric M, Sopta J, Djonc D, Lujic N. Skeletal manifestations of hydatid disease in Serbia: demographic distribution, site involvement, radiological findings, and complications. Korean J Parasitol 2013;51:453-9.
7. Gun E, Etit D, Buyuktalanci DO, Cakalagaoglu F. Unusual locations of hydatid disease: A 10-year experience from a tertiary reference center in Western Turkey. Ann Diagn Pathol 2017;29:37-40.
8. Vasilevska V, Zafirovski G, Kirjas N, Janevska V, Samardziski M, Kostadinova-Kunovska S, et al. Imaging diagnosis of musculoskeletal hydatid disease. Prilozi 2007;28:199-209.
9. Pedrosa I, Saiz A, Arrazola J, Ferreiros J, Pedrosa CS. Hydatid disease: radiologic and pathologic features and complications. Radiographics 2000;20:795-817.
10. Tüzün M, Hekimoğlu B. CT findings in skeletal cystic echinococcosis. Acta Radiol 2002;43:533-8.
11. Arazi M, Erikoglu M, Odev K, Memik R, Ozdemir M. Primary *Echinococcus* infestation of the bone and muscles. Clin Orthop Relat Res 2005;234-41.
12. Aslan B, Aslan G, Ozardali I. Multilocular echinococcosis in humerus: a case report. J Egypt Soc Parasitol 2001;31:961-2.
13. Chari PR. Hydatid disease of scapula and upper third of humerus treated by en bloc excision and fibular bone grafting. Indian J Orthop 2007;41:241-3.
14. Markonis A, Tavernarakis A, Papaevangelou M. Humeral hydatid cyst complicated with extrasosseous involvement: a case of unusual location of echinococcosis. Eur J Radiol 2001;37:130-3.
15. Nourbakhsh M, Shemshaki H, Zarezadeh A, Etemadifar MR, Mazoochian F. Recurrent hydatidosis at the site of non-union humerus fracture. Int J Prev Med 2012;3:660-3.
16. Ozkan H, Dogramaci Y, Kose O, Esen E, Erdem H, Komurcu M. Primary hydatid disease of the humerus. Ann Acad Med Singapore 2008;37:440-1.
17. Laubscher M, Hardcastle PS, Naude PH, Barrett TN. Hydatid disease of bone. SA Orthop J 2008;Winter:44-7.
18. Eckert J, Deplazes P. Biological, epidemiological, and clinical aspects of echinococcosis, a zoonosis of increasing concern. Clin Microbiol Rev 2004;17:107-35.
19. Gottstein B, Reichen J. Echinococcosis/hydatidosis. In: Cook GC, Zumla AI, editors. Manson's tropical diseases. 22nd ed. Philadelphia: Saunders Elsevier; 2009. p. 1549-68.
20. Taljanovic MS. Atypical infections. In: Pope TL Jr, Bloem HL, Beltran J, Morrison WB, Wilson DJ, editors. Musculoskeletal imaging. 2nd ed. Philadelphia, PA: Elsevier Saunders; 2015. p. 848-51.
21. Bullough PG. Bone and joint infection. In: Bullough PG, editor. Orthopaedic pathology. 5th ed. St. Louis: Mosby/Elsevier; 2010. p. 134-9.