

A Rare Case of Human *Dioctophyma renale* infection in an Adult Filipino Male

Johanne S. Baluyut, MD,¹ Edison Guerrero, MD,¹ Paolo Villareal, MD¹

Abstract

Introduction: *Dioctophyma renale*, also known as the “giant kidney” worm is the largest nematode that infects mammals. Most commonly, it infects dogs and only a few cases of human infestation have been reported.

Case Presentation: In this case, we present a 71-year-old male with a history of right flank pain, painless hematuria, obstructive urinary symptoms and occasional passage of a long red meat-like structure via urethra. Previous ultrasonography revealed a renal mass that was interpreted as renal malignancy. Patient was advised radical nephrectomy with biopsy; however, he was lost to follow up. After 8 years, he again presented with a 2-month history of right flank pain, gross hematuria, unintentional weight loss and obstructive urinary symptoms. He was admitted to our institution and laboratory findings revealed microcytic hypochromic anemia. Ultrasound showed a focal bulge in the superior wall of the urinary bladder and an enlarged right kidney with a large heterogeneous solid structure. Insertion of a foley catheter relieved the urinary obstruction and there was noted passage of a single roundworm measuring 55 cm by 0.32 cm. The specimen was sent for histopathology and microscopic examination revealed a necrotic body of a roundworm consistent with *Dioctophyma renale*.

Case Discussion: The clinical manifestations of *D. renale* infection are non-specific and the diagnosis can be a challenge as it commonly mimics renal malignancy and other intra-abdominal tumors. The diagnosis is often an incidental finding of the parasite on necropsy or by finding ova in the urine. The clinical findings of painless hematuria, unintentional weight loss and an ultrasound finding of renal mass usually point toward the diagnosis of renal cell carcinoma.

As we present this case, it will be highlighted that such signs and symptoms, such as painless hematuria, unintentional weight loss and ultrasound finding of a renal mass, can also be found in patients with rare human parasitism from *Dioctophyma renale*.

Keywords: parasite, nematode, *Dioctophyma*, renal mass, giant, worm, carcinoma, kidney

Introduction

Dioctophyma renale also known as the “giant kidney worm” is the largest known parasitic nematode belonging to the order of *Ascaridia* and family of *Dioctophymatidae*. A German zoologist, Johann Goez, first described it in 1782 after discovering the worm in a dog’s kidney^{5, 18}. It inhabits temperate regions worldwide, particularly areas with freshwater streams and lakes. It has a predilection to infect the kidneys of a variety of fish-eating mammalian species, such as dog, wolf, cheetah, coyote, fox, mink, horse, and swine and transmission is believed to be by consumption of raw fish^{4, 6, 9}. Mammals are the definitive host of the nematode

as it prefers to complete its life cycle in the mammalian kidney, preferentially on the right side^{3, 11}. Human infestation is rare, first of which was documented in 1845 in an Iranian female who presented only with nonspecific symptoms of fever and laboratory finding of eosinophilia¹². A review of related literature revealed less than 20 cases of human *Dioctophyma renale* infection to date^{1, 2, 7, 10, 12, 14-18, 21, 24-31}. *D. renale* has been found in both right and left kidneys, skin, subcutaneous nodules, retroperitoneum, and the liver^{1, 2, 25, 26}. This case is unique as it presents both as a renal and urinary bladder mass and is the first documented case of *D. renale* infection in the Philippines.

Case Report

We present a case of a 71-year-old Filipino male with an eight-year history of right flank pain, painless hematuria and obstructive symptoms relieved by intermittent insertion of foley catheter with several episodes of

¹ Department of Internal Medicine, James L Gordon Memorial Hospital

Corresponding author: Johanne S. Baluyut, MD
Email: hanne_jho676@yahoo.com

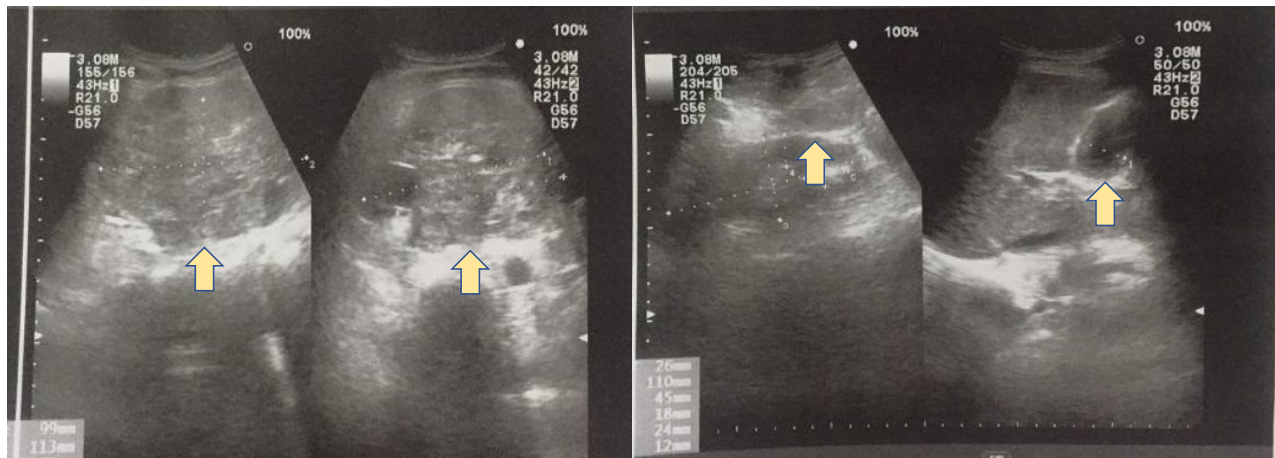


Figure 1 Ultrasound of the right kidney showing an enlarged right kidney measuring 16.4 x 8.5 x 10.0 cm with large heterogenous solid structure measuring 11.3 x 7.7 x 9 cm (left picture) and a focal bulge in the superior wall of the urinary bladder to consider mass versus cystitis glandularis (right picture).

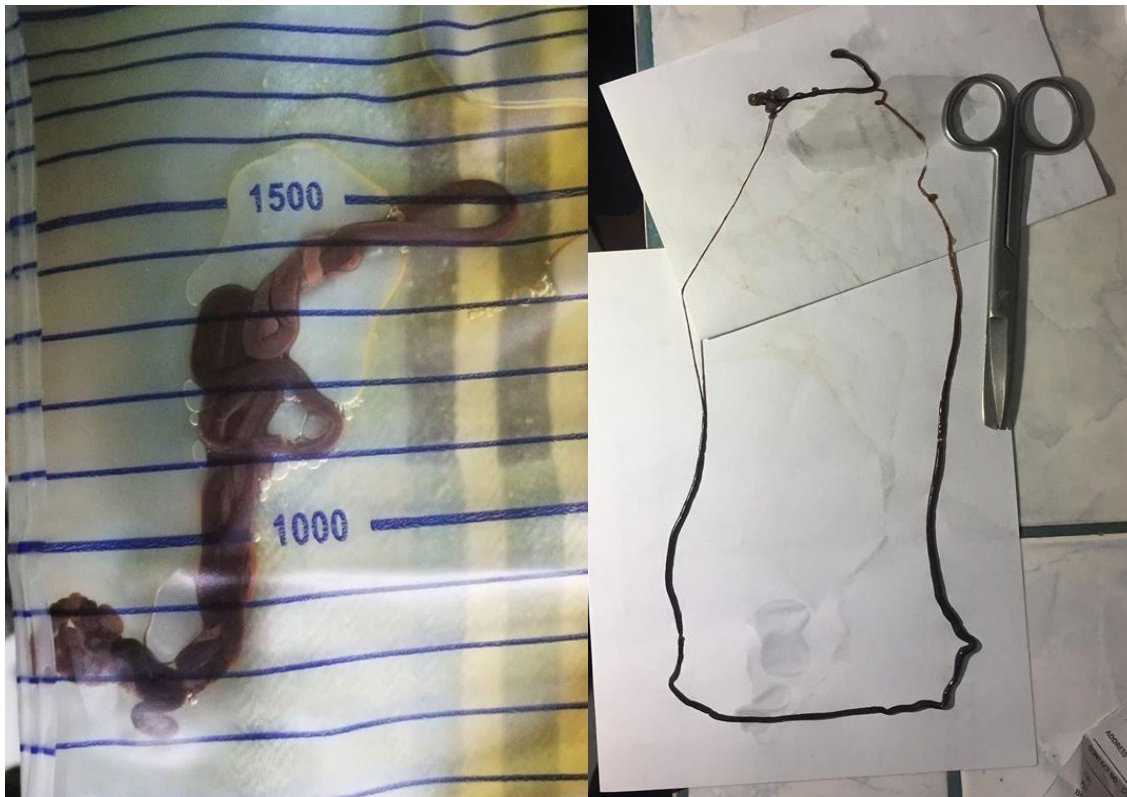


Figure 2 An adult *Dioctophyma renale* seen in the patient's urine bag (left picture) and Photograph of a giant kidney worm single roundworm measuring 55 cm in length and 0.3 cm in diameter (right picture).

spontaneous passage of elongated red tissue per urethra.

The patient was noted to be fond of eating raw fish and occasionally raw dog meat marinated in lime and other spices. Ultrasound and computed tomography revealed a solitary right renal mass interpreted by his previous physician as renal malignancy. The patient was then

advised radical nephrectomy with biopsy; however, he was lost to follow up. After 8 years, the patient again presented with a 2-month history of right flank pain, painless gross hematuria, unintentional weight loss, pallor and obstructive urinary symptoms.

On physical examination, the patient was pale and was noted to have muscle and fat wasting. He had a soft, non-

tender abdomen, positive right costovertebral angle tenderness and no palpable mass. He was tachycardic but with otherwise normal vital signs. Laboratory findings revealed hemoglobin of 7.2 g/dL, WBC $8.82 \times 10^9/L$, thrombocytes 745,000, blood urea nitrogen 7.27 mmol/L, creatinine 142.28 $\mu\text{mol/L}$, PSA 2.56 ng/ml, urine sediments WBC 4-6/hpf, RBC >30/hpf; no egg of worms was found in the urine and urine cytology was negative for malignant cells. His anemia was corrected by transfusion of three-unit packed RBC. In order to rule out a renal tumor which could explain the patient's anemia, painless hematuria and flank pain, a kidney ultrasound was requested. Ultrasound showed a focal bulge in the superior wall of the urinary bladder to consider mass versus cystitis glandularis, a normal sized prostate and an enlarged right kidney with large heterogeneous solid structure. (Figure 1).

On the 4th hospital day, there was noted spontaneous passage of a single roundworm in the patient's urine bag. (Figure 2). The specimen was sent for histopathology and microscopic examination revealed a gangrenous necrotic body of a roundworm consistent with *Diectophyma renale*. The patient was advised to do a CT scan with contrast of the whole abdomen to delineate the renal mass, its relationship to the adjacent organs and to determine its resectability. Patient refused surgical intervention and requested to be discharged after correction of his anemia. He was advised for outpatient follow up.

Discussion

Diectophyma renale (giant kidney worm) is one of the largest parasitic nematodes that infects carnivorous mammals, such as minks, canids, dogs, and cats^{8, 13, 21, 24}. *D. renale* was first reported by Goez in 1782 in the canine kidney^{5, 18}. Adult male worms measure up to 35 cm x 3-6 mm wide, and females up to 103 cm x 5-12 mm wide. Both sexes are blood red in color with a round body and blunt slightly tapered ends. The worms usually occur in the kidneys and more frequently in the right organ than in the left^{6, 9, 24}. The eggs are voided in urine and first-stage larvae develop in 15-100 days. Embryonated eggs are ingested by aquatic intermediate hosts, usually by an aquatic worm. Within the intermediate host, the larvae molt twice and become infectious. The hosts are exposed to the parasite by ingesting encysted larvae in raw fish (eg, pike, bullhead) or frogs, or by ingesting an infected annelid worm. The larvae penetrate the bowel wall and migrate first to the liver. Juvenile larvae mature over a period of about 50 days in the liver parenchyma, and then migrate directly to the kidneys to mature further; they may remain alive for up to 5 years. The worm(s) can cause obstruction, hydronephrosis, and destruction of the renal parenchyma^{3, 11, 20}.

Human infection is rare and was first reported in 1845 in a female Iranian who initially presented with fever and eosinophilia. Further work up noted the presence of *D. renale* ova in the urine. However, renal angiography, and retrograde pyelography, did not reveal the location of the parasite¹². These were probably negative because

these modalities do not evaluate the renal parenchyma which the parasite usually affects. In contrast our patient underwent ultrasonography which revealed a large heterogeneous solid structure in the renal parenchyma.

Since then, there have been several reports of < 20 human infestation and more often than not, have been found accidentally during necropsy or biopsy.

The clinical manifestations of *D. renale* infection are non-specific. If the parasite lodges in the urethral meatus it can cause obstructive symptoms and as the parasite destroys the renal parenchyma, *D. renale* usually presents with hematuria, which if persistent can lead to anemia, and a renal mass with or without flank pain as what was seen in our patient. The diagnosis can be a challenge as it mimics not only renal malignancy^{10, 15, 16} but also other intra-abdominal and retroperitoneal tumors as well^{23, 25, 26, 32}. One case report from Korea presented with *D. renale* infection occurring simultaneously with malignant fibrous histiocytoma of the ureter²⁵ and another case reported *D. renale* infection mimicking a retroperitoneal neoplasm²⁶. Only a few parasites have been documented to infect the genitourinary system. *D. renale* is unique because it causes destruction by consuming the renal parenchyma, causing hematuria and obstructive symptoms, and sparing the capsule^{7, 16, 29}.

At present, surgical management by excision of the tumor remains the recommended treatment that has positive effect on long term survival of patients infected with *D. renale*. Nephrectomy is considered for severe cases with extensive involvement of the renal parenchyma¹⁴. The use of anti-helminthic drugs has yet to be evaluated as a conservative approach to treat diectophymiasis^{10, 16, 29}. However, three cases recorded in China who received albendazole later observed no passage of adult worms and eggs after treatment^{19, 32, 33}. While in another case report, ivermectin effectively cured a patient after two courses of this medication¹⁴.

Conclusion

In this case report, we are presented with a 71-year-old male with right lumbago, painless gross hematuria, anemia, unintentional weight loss and obstructive urinary symptoms similar to the clinical presentation of all the other reported cases. Together with an ultrasound finding of a renal and urinary bladder mass, it is common to misdiagnose this as a case of renal malignancy with probably bladder extension. However, taking into account the history of intake of raw fish and the patient's temporal profile, that despite the chronicity of his disease, the patient had only presented with such benign symptoms, *D. renale* infection should be considered as a differential diagnosis still with the intention to rule out the presence of any possible concomitant malignancy. In addition to the rarity of the case, this is the first documented human infection of *D. renale* in the Philippines.

References

1. Beaver PC, Theis JH. Diectophymatid larval nematode in a subcutaneous nodule from man in California. *Am J Trop Med Hyg.* 1979; 28: 206-12.
2. Beaver, P. C. and C. Khamboonruant. 1984. *Diectophyma*-like larval nematode in a subcutaneous nodule from man in northern Thailand. *American Journal of Tropical Medicine and Hygiene* 33:1032-1034.
3. Centers for Disease Control and Prevention. DPDx - Laboratory Identification of Parasitic Diseases of Public Health Concern: Diectophymiasis. <http://www.cdc.gov/dpdx/diectophymiasis/index.html> (accessed 16 Jul 2015).
4. Celerin, A. J. and M. E. McMullen. 1981. Giant kidney worm in a dog. *Journal of the American Veterinary Medical Association* 179:245-246.
5. Crichton VJ, Urban RE, *Diectophyma renale* (Goeze, 1782) (Nematoda: Diectophymata) in Manitoba mink Canadian Journal of Zoology 1970 May;48(3):591-2
6. *Diectophyma renale* [cited 2006 July 17]. Available from: <http://www.stanford.edu/class/humbo103/ParaSites>.
7. Fernando SS. The giant kidney worm (*Diectophyma renale*) infection in man in Australia. *Am J SurgPathol* 1983;7:281-4.
8. Gargili A, Firat I, Toparlak M, Çetinkaya H. First case report of *Diectophyma renale* (Goeze, 1782) in a dog in Istanbul, Turkey. *Turk J Vet Anim Sci.* 2002;26:1189-91.
9. Griffiths HJ. A handbook of veterinary parasitology. Minneapolis: University of Minnesota Press; 1978.
10. Gu Y, Li G, Zhang J, et al. *Diectophyma renale* infection masquerading as a malignancy. *Kidney Int* 2012;82:1342.
11. Hallberg, C. W. 1953. *Diectophyma renale* (Goeze, 1782). A study of the migration routes to the kidneys of mammals and resultant pathology. *Transactions of the American Microscopical Society* 72:351-363.
12. Hanjani AA, Sadighian A, Nikakhtar B, et al. The first report of human infection with *Diectophyma renale* in Iran. *Trans R Soc Trop Med Hyg* 1968;62:647-8.
13. Hoffman V, Nolan TJ, Schoelkopf R. First report of the giant kidney worm (*Diectophyma renale*) in a harbor seal (*Phocavitulina*). *J Parasitol.* 2004;90:659-60.
14. Ignjatovic I, Stojkovic I, Kutlesic C, et al. Infestation of the human kidney with *Diectophyma renale*. *UrolInt* 2003;70:70-3.
15. Katafigiotis I, Fragkiadis E, Pournaras C, et al. A rare case of a 39 year old Male with a parasite called *Diectophyma renale* mimicking renal cancer at the computed tomography of the right kidney. A case report. *ParasitolInt* 2013;62: 459-60.
16. Kuehn J, Lombardo L, Janda WM, et al. *BMJ Case Rep* Published online: [25 January 2016] doi:10.1136/bcr-2015-212118
17. Lei B, Pang YQ, Kong BQ. Report on a case of *Diectophyma renale* infection. *Chin J Parasitol Parasitic Dis.* 2002;20:151.
18. Li G, Liu C, Li F, et al. Fatal bilateral diectophymatosis. *J Parasitol* 2010;96:1152-4.
19. Liu DX. 2001. The first finding of human infection with *Diectophyma renale* from Heilongjiang Province, China. *Chinese Journal of Parasitic Disease Control*, 14(1), 80 (in Chinese).
20. Mace, T. F. and R. C. Anderson. 1975. Development of the giant kidney worm, *Diectophyma renale* (Goeze, 1782) (Nematoda: Diectophymatoidea). *Canadian Journal of Zoology-Revue Canadienne de Zoologie*53:1552-1568.
21. Measures, L. N. 2001. Diectophymatosis. In Parasitic diseases of wild animals. Samuel, W. M., M. J. Pybus, and A. A. Kocan. (eds.). Iowa State University Press. Ames, Iowa. p. 357-364.
22. Narvaez, J. A., L. P. Turell, and J. Serra. 1994. Hyperdense renal cystic lesions caused by *D. renale*. *American Journal of Roentgenology and Radium Therapy* 163:997-998.
23. Narvaez, J. A., L. P. Turell, and J. Serra. 1994. Hyperdense renal cystic lesions caused by *D. renale*. *American Journal of Roentgenology and Radium Therapy* 163:997-998.
24. Palmer PES, Reeder MM. Kidney worm: Diectophymiasis and eustrongylidiasis. In: The imaging of tropical diseases, with epidemiological, pathologic, and clinical correlation chapter 46. The Uniformed Services University of the Health Sciences; 2007. Available from: <http://tmcr.usuhs.mil/tmcr/toc.htm>
25. Park HY, Seo JW, Lee BH, et al. Simultaneous occurrence of malignant fibrous histiocytoma of the ureter and *Diectophyma renale* infection: a case report. *J Korean SocRadiol* 2013;68:411-15.
26. Sun T, Turnbull A, Lieberman PH, et al. Giant kidney worm (*Diectophyma renale*) infection mimicking retroperitoneal neoplasm. *Am J SurgPathol* 1986;10:508-12.
27. Tokiwa T, Ueda W, Takatsuka S, et al. The first genetically confirmed case of *Diectophyma renale* (Nematoda: Diectophymatida) in a patient with a subcutaneous nodule. *Parasitol Int* 2014;63:143-7.
28. Urano, Z., H. Hasegawa, and T. Katsumata. 2001. Diectophymatid nematode larva found from human skin with creeping eruption. *Journal of Parasitology* 87:462-465.
29. Venkatrajaiah N, Kalbande SH, Rao GV, et al. Diectophymatosis renalis in humans: first case report from India. *J Assoc Physicians India* 2014;62:70-3.
30. Vibe, P. P. 1985. *Diectophyma* infection in humans. *Meditinskaiaparazitologiiaparazitarnyebolozni (Mosk)* 1:83-84.
31. Vladimova, M. G., Ala Lysenko, and lu P. Gorbunova. 2002. A case of diectophymosis (*Diectophyma renale*) in a girl from Arkhangelsk. *Meditinskaiaparazitologiiaparazitarnyebolozni (Mosk)* 4:48-50.
32. Yang, J., Li, P., Su C., YiZhang, J., and Gu, M. 2016. Worms Expelled With the Urine From a Bosniak Cyst III of the Left Kidney. *Urology* 93: e5.
33. Yang YR, Lu YY. 1995. A case report of diectophymiasis infection cured by albendazole. *Chinese Journal of Parasitology and Parasitic Diseases*, 13(13), 192 (in Chinese).