CASE REPORT

HUMAN PENTASTOMIASIS IN UPPER EAST REGION OF GHANA: REPORT OF TWO CASES

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Abstract -

Objective: The prevalence of human pentastomiasis in the North-Eastern and Upper East Region of Ghana is probably high since numerous incidental cases have been reported from there. This paper reports two new cases of human pentastomiasis diagnosed incidentally at a District Hospital in the Upper East Region of Ghana, discusses the diseases that the infestation cause and stresses the public health issues this infestation brings up.

Case Reports: The parasites were diagnosed incidentally in two males presenting for elective inguinal herniorrhaphy. Two encysted larvae were in the hernia sacs of two normal males, who were operated

upon at the Sandema District Hospital in February 2009 during one of the annual charity surgical outreach services in that hospital. The worms were dissected out and identified morphologically as *Armillifer armillatus*, based on prior knowledge from our previous experience in diagnosing a case in our institution both from histopathological examination and confirmation by a medical parasitologist.

Conclusion This curious but important zoonotic parasitic infestation should be of public health concern since the *Armillifer armillatus* which infests man in West Africa also infests dogs, one of man's closest pets.

Key Words: Intestinal Parasites, Python, Totemism, Upper East Region, Ghana.

Introduction

A neglected and little known zoonotic parasitic infestation, human pentastomiasis, is repeatedly reported from the Upper East Region of Ghana. Two Army recruits from Paga and Bawku in the North-Eastern Area of Ghana, were diagnosed radiologically as having human pentastomiasis in 1962 at the 37 Military Hospital by Lt Col P. M Betland¹. Thereafter, there was a void in the diagnosis of this parasite in Ghana until one was encountered accidentally at Korle Bu Teaching Hospital in 2006². Following this three other cases were diagnosed in people, all from the North-Eastern and Upper East Regions of Ghana, prompting renewed interest in this parasite. These cases were investigated and it was found that all of them had had close association with the python. This association has led to direct transmission of this parasite to humans in Ghana and explains the increasing number of cases from this particular area of Ghana³.

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Author for Correspondence: Dr Jonathan CB Dakubo Department of Surgery University of Ghana Medical School Korle Bu. E mail: jdakubo@yahoo.com Conflict of interest: None declared Resource Foundation (CMRF), who visit Sandema District Hospital, in the Builsa district of the Upper East Region of Ghana annually to offer surgical service to the people. An incidental finding of two new cases of human infestation with this parasite at Sendema District Hospital forms the basis of this report.

Cases Reports

The two cases, both males, presented with uncomplicated huge inguinal hernia to the Sendema District hospital in February 2009 when message of the charity surgical outreach programme got to them. Case 1 was a 35-year old man who came from Fumbisi and Case 2, a 53-year old man from Wiaga all in the Upper East region of Ghana. They did not have symptoms referable to the gastrointestal tract nor did they have any cough or chest pains. They were both emaciated but had stable cardiovascular and respiratory system activities. They both had huge left inguinal hernia with wide necks which made the hernia easily reducible. The haemoglobin of Case 1 was 11.8g/dl and that of Cases 2 was 10.2g/dl. Stool microscopy did not yield any parasites. Blood urea, electrolytes and creatinine were not done. They both underwent tension-free nylon darn repair of the hernia under spinal anaesthesia. The sacs of both hernias contained viable small intestine. Encysted pentastomid larvae (Figs. 1and 2) were found in the hernia sacs. They were excised and dissected for morphological identification after surgery. The identification of the parasites as Armillifer armilatus was based on knowledge acquired by the authors on

this parasite when a case was diagnosed earlier both by histopathological examination and confirmation by a medical parasitologist². Also, other cases have been identified by the first author and a pathologist ³. Plain chest and abdominal X-rays were taken but no calcified lesions were identified. The patients were informed about the parasitic infestation and reassured of no complications from the infestation since it was indolent in the absence of symptoms. They admitted to the python being their totem which forbade its consumption. They were discharged on medication including albendazole.



Fig 1. Pentastomid in excised hernia sac from case one. Parasite is shown by arrow head



Fig 2. Larva of pentastomid from case one measures 6mm in diameter

Discussion

Fumbisi and Wiaga are near to Sandema where these people sojourned to access health care. They originally came from very remote communities in the South-Western part of the Upper East Region extending into the mid-upper part of the Northern Region. These communities are not easily accessible and referred to

as 'overseas'. These communities are close to the Mole Game reserve on its Eastern border. Conditions there are still quite primitive and cohabitation of humans and animals, including reptiles still takes place. Pentastomiasis is a zoonotic infestation of humans. The adult worms dwell in the respiratory passages of the python⁴. Humans are accidentally infested creating a cul-de-sac in the chain of transmission. Other mammals such as; dogs, sheep, cattle and in the wild, both herbivores and carnivores are equally infested⁵⁻⁹. The number of reported cases of human pentastomiasis in the world literature is increasing, with many coming from West and Central Africa $^{10-12}$. Reported cases in the high income nations are usually immigrants from the tropics and subtropics 5,13,14. Some cases are also reported from Asia where snake meat is consumed¹⁵. Consumption of inadequately cooked snake meat, contact with excretions from snakes, particularly the python as in a totemic relationship and unscientific snake farming are the major circumstances through which transmission to humans can occur^{16,17}. Chance transmission of the embryonated eggs to humans from contaminated vegetation and water is exceedingly rare and cannot explain the clustering of cases or the massive infestations being reported in the literature.

The two cases presented here responded to an advertisement announcing the coming of a team to offer specialist surgical service in their community. Besides their hernia they were in apparent good health and infestation with pentastomids was the least likely diagnosis that one could envision, let alone identifying the larvae in a hernia sac. Human Pentastomiasis is usually an indolent infestation and only infrequently a cause of disease. It is mostly diagnosed incidentally during surgery as in these two cases, at autopsy or on plain radiographs of the abdomen and chest taken for some other reason ¹. To date nearly all reported cases of this parasitic infestation in humans in Ghana have been from the Upper East Region and North Eastern parts of Ghana.

It has been shown that totemism plays a role in the transmission of this parasite from the python to man in Ghana. These two cases come from the same region of Ghana where earlier cases of the parasite have been reported. They come to add to the tally making a total of seven out of the eight known cases coming from this area since the first report was made at the 37 Military Hospital in 1962. Housing and other services have improved recently in the Upper East Region of Ghana and the expectation should be that reptiles, like the python, will sparingly creep into human habitats. However there are still communities where people still live in shelters that are insecure for human habitation. These two cases came from 'overseas' where human beings still co-habit with animals, including reptiles. The communities are still primitive and social amenities and living conditions there are still far from acceptable. Considerable public health education together with social interventions are required to address transmission of infectious diseases, including pentastomiasis.

Significant disease may develops in cases of massive infestation that could be fatal. But such cases

may go unnoticed in a setting where post-mortem examinations are not done on the dead.

Ingested embryonated eggs hatch in the small intestine and moult into first stage larvae. They migrate through the intestinal wall. Entry into a submusocal venule results in transportation of the larve into a hepatic portal vein where the larvae might reside until they perish.³ Parasites can migrate through the substance of the liver to encyst below the Glisson capsule. Spread through the intestinal lymphatics occur. Such lymphatic spread transports the parasites into the lungs and the hilar lymph nodes. The lack of reports of the parasites recovered from muscle, bone and skin could suggest that systemic haematogenous spread is halted by filtration of the parasites in the lung they have drained into the parenchyma after innominate vein from the left thoracic duct. Two reports, one of the brain and the other in the eye however, may tend to contradicts this assertion¹⁸⁻²⁰. The intraocular infestation could however be from direct innoculation.

Complete transmural intestinal migration brings the parasites, usually measuring about 250µm, to the subserosal space where they encyst or are shed into the peritoneal cavity and spread to other places for encystment. They encyst in the omentum, mesentery and the parietal peritoneum from where they could be identified in the hernia sac, as in these two cases. The mechanism by which the transmural migration occurs has not been defined, but could be by enzymatic digestion of the bowel wall by the lead point of the larva, and as it moves through the bowel wall, healing takes place from behind²¹. Massive simultaneous transmural intestinal migration of the larvae or their lodgement in mesenteric lymph nodes causes gastrointestinal symptoms, particularly severe abdominal cramps^{22,23}. On occasion when many parasites are migrating through the bowel wall or massive mesenteric lymph node infestation occurs, extensive tissue damage resulting in bowel necrosis and gangrene or proliferative generalised peritonitis develops^{24,25}. Not uncommonly extensive peritoneal adhesion formation can occur as a result of reaction to the parasites. The adhesions could then cause mechanical intestinal obstruction²⁶.

Not all the parasites which hatch in the intestinal lumen possess the capacity for transmural migration, a phenomenon needing evaluation. Those unable to migrate reside in the lumen of the intestine and anchor themselves to the mucosa with their five hooks and feed on blood therefrom. They are shed in the faeces spontaneously or following treatment with an antihelminthic, such as albendazole^{15,22,26}. Heavy intraluminal parasitisation causes severe nutritional problems, anaemia, and weight loss²². A combined massive intra- and extra-luminal parasitic infestation is often fatal, be it in humans or animals²⁷⁻³⁰.

Intraperitoneal parasites remain encysted as long as the person or the infested mammal is alive. They moult into third stage larvae and remain encysted till they perish after several years and then undergo dystrophic calcification. In humans and other large marmals this ends the life cycle as the larvae are incapable of further migration through the bowel wall.The calcified parasites are seen on plain abdominal or chest X-Ray as circular or C-shaped opacities. Reported cases of free motile parasites in the peritoneal cavity are from autopsy specimens only³. What triggers the parasites to excyst following the death of the host is unclear. A sudden drop in the temperature of the host and/or cessation of nutrient flow (blood) to the parasite could account for this phenomenon, but these need investigation to confirm.

The macroscopic size of the encysted parasite cannot be missed when it is encountered in the peritoneum at laparotomy or at autopsy. They are striking in appearance. The diameter of the curled up larvae of *Armillifer armillatus* measure on average, 0.5-0.6cm (fig. 1b) and the free larva about 1.9-2.3cm.17 The encysted larvae are characteristically curved (figure 1) and covered by a thin transparent membrane and remain in this curled up position and feed on blood as long as the host is alive.

The identification of the parasites as *Armillifer armillatus* in these two cases was based solely on their morphological features and their similarity to those previously published from Ghana. They possessed the same features as the earlier ones measuring 2.0cm in length when dissected out. Detailed characterization of the parasites using the novel Polymerase chain reaction is the ideal, but this was not available to us at the time the parasites were diagnosed.

The other species of pentastomids that infest human beings are *Armillifer grandis*, which together with *Armillifer armillatus* are found in Africa^{2,3,5}. *Armillifer agkistrodonti* is predominantly found in China³¹ and *Armillifer moniliformis* in Southeastern Asia¹⁰.

There is no literature on drug treatment for this parasite. Albendazole interrupts glucose utilization in the cells of the adult and larval forms of many intestinal parasites leading to energy depletion and death, and has been used effectively against a wide variety of intestinal worms of different species. With reports showing its capability of expelling pentastomids from the gastrointestinal tract, it is worth trying in massive infestations showing significant symptoms. Otherwise the infestation should be left alone since the parasite will die naturally after about two years.

Conclusion

To date all known cases of human pentastomiaisis in Ghana have been diagnosed incidentally. These undoubtedly could represent a small portion of an epidemiologically significant disease that has not been pursued. There is an urgent need to determine the true prevalence of infestation by this parasite in the NorthEastern and Upper East Region of Ghana using serological screening⁵. Human pentastomiasis is therefore of public health concern granting that transmission of the same species of the parasite, *Armillifer armillatus*, to dogs and to humans has been reported^{5,24}.

References

- 1. Bretland PM. Armillifer armillatus infection: Radiological diagnosis in two Ghanian soldiers. *Br. J. Radiology*. 1962; 35: 602-608
- 2. Dakubo JCB, Etwire VK, Kumoji R, Naaeder SB. Human Pentastomiasis: A case report. West *African J. of Med.* 2006; 25: 166-168.
- 3. Dakubo JCB, Naaeder SB, Kumoji R. Totemism and the transmission of human pentastomiasis. *Ghana Med J.* 2008; 42: 165-168
- 4. Ayinmodei AB, Adedokun AO, Aina A, Taiwo V. The zoonotic implications of pentastomiasis in the royal python. Ghana Med J. 2010; 44: 115-118.
- 5. Tappe D, Meyer M, Oesterlein A, Jaye A, Frosch M, et al. Transmission of Armillifer armillatus Ova at Snake Farm, The Gambia, West Africa. *Emerging Infectious Diseases*. 2011; 17: 251-254
- 6. Brookins MD, Wellehan JF, Roberts JF, Allison K, Curran SS, Childress AL, Greiner EC. Massive visceral pentastomiasis caused by Porocephalus crotali in a dog. *Vet Pathol.* 2009; 46: 460-463.
- Oryan A, Khordadmehr M, Ranjbar VR.Prevalence, biology, pathology, and public health importance of linguatulosis of camel in Iran. *Trop Anim Health Prod.* 2011; 43: 1225-1231.
- 8. Nourollahi Fard SR, Kheirandish R, Asl EN, Fathi S. Mesenteric and mediastinal lymph node infection with Linguatula serrata nymphs in sheep slaughtered in Kerman slaughterhouse, southeast Iran. *Trop Anim Health Prod*. 2011; 43: 1-3.
- Young E. Pentastomiasis (Armillifer and Linguatula Sp.) infestations of wild animals in the Kruger National Park. J S Afr Vet Assoc. 1975; 46: 335-336.
- Yao MH , Wu F , Tang LF , 2008 . Human pentastomiasis in China: case report and literature review. *J Parasitol*. 2008; 94: 1295 – 1298.
- 11. Burns-Cox CJ , Prathap K , Clark E , Gillman R. Porocephaliasis in western Malaysia . *Trans R Soc Trop Med Hyg.* 1969; 3: 409 – 411.
- Prathap K , Lau KS , Bolton JM. Pentastomiasis: a common finding at autopsy among Malaysian Aborigines. *Am J Trop Med Hyg.* 1969; 18: 20 – 27.
- 13. du Plessis V, Birnie AJ, Eloff I, Reuter H, Andronikou S. Pentastomiasis (Armillifer armillatus infestation). *SAMJ*. 2007; 97: 928-930.
- Guardia SN, Sepp H, Scholten T, Morava-Protzner I. Pentastomiasis in Canada. *Arch Pathol Lab Med.* 1991; 115: 515-517.
- 15. Ye F, Sheng Z-K, Li J-J, Sheng J-F. Severe

pentastomiasis in children: A report of 2 cases. Southeast Asian J TropMed PublicHealth. 2013; 44: 25-30.

- Rail GA. Porocephaliasis: a description of two cases in Sabah. *Trans R Soc Trop Med Hyg.* 1967; 61: 715 – 717.
- 17. Tappe D, Büttner DW, 2009. Diagnosis of human visceral pentastomiasis. *PLoS Negl Trop Dis.* 2009; 3: 320-321.
- Cagnard V, Nicolas-Randegger J, Dago Akribi A, Rain B, et al. Generalized and lethal pentastomiasis due to Armillifer grandis (Hett, 1915). *Bull Soc Pathol Exot Filales*. 1979; 72: 345-352.
- 19. Pal SS, Bhargava M, Kumar A, Mahajan N, et al. An unusual intraocular tongue worm in anterior chamber: a case report. *Ocul Immunol Inflamm*. 2011; 19: 442-443.
- Fontanel A, Doucet J, Loubiére R. Conjunctival pentastomiasis. Case report. *Nouv Presse Med.* 1972; 1: 3123-3124.
- 21. Jones DA, Riley J, Kerby NW, Knox DP. Isolation and preliminary characterization of a 48-kilodalton metalloproteinase from the excretory/secretory components of the frontal glands of Porocephalus pentastomids. *Mol Biochem Parasitol*. 1991; 46: 61-72.
- 22. Wang H-Y, Zhu G-H, Luo S-S, Jiang K-W. Childhood pentastomiasis: A report of three cases with the following-up data. *Parasitology International*. 2013; 62:289-292
- 23. Herzog U, Marty P, Zak F. Pentastomiasis: case report of an acute abdominal emergency. *Acta Trop.* 1985; 42: 261-271.
- 24. Stief B, Enge A. Proliferative peritonitis with larval and cystic parasitic stages in a dog. *Vet Pathol.* 2011; 48: 911-914
- 25. Ma KC, Qiu MH, Rong YL. Pathological differentiation of suspected cases of pentastomiasis in China. *Trop Med Int Health*. 2002; 7: 166-177
- 26. Qiu MH, Ma KC, Fan PC, Lu SS. Discovery of a new species of the pentastomid genus Porocephalus (Humboldt, 1811) from Taiwan, China and its pathogenic features. *Zhonqquo Ji Sheng Chong Xue Yu Ji Sheng Chong Bing Za Ahi*. 2005; 23: 69-72.
- 27. Tiendrebeogo H, Levy D, Schmidt D.Human pentastomiasis in Abidjan: A report on 29 cases. *Rev. Fr Mal Respir.* 1982; 10: 351-358.
- Lavarde V, Fornes P. Lethal infection due to Armillifer armillatus (Porocephalida): a snakerelated parasitic disease. *Clin Infect Dis.* 1999; 29: 1346–1347.
- 29. Adams L, Isaza R, Greiner E. Fatal Pentastomiasis in a captive African dwarf crocodile hatchlings (Osteolaemus tetraspis). *J Zoo Wildlife Med*. 2001; 32: 500-502.

- Yapo Ette H, Fanton L, Adou Bryn KD, Botti K, Koffi K, Malicier D. Human pentastomiasis discovered postmortem *Forensic Sci Inter*. 2003; 137: 52–54.
- 31. Chen SH, Liu Q, Zhang YN, Chen JX, Li H,

Chen Y, Steinmann P, Zhou XN. Multi-host model-based identification of Armillifer agkistrodontis (Pentastomida), a new zoonotic parasite from China . *PLoS Negl Trop Dis* 2010; 4: e647 .