

Scrotal Paragonimiasis in adults

Two case reports and review of literature

Tiancai Liang, MS, Guobiao Liang, MD*, Yang Du, MS, Xin Wang, MS, Yuanliang Wang, MS, Anjian Chen, MS, Zongping Chen, MD, Jiang Du, MS, Hao Li, MS, Lang Yu, MS

Abstract

Rationale: Paragonimiasis is a parasitic disease caused by *Paragonimus* in the lungs; it can be divided into intrapulmonary type and extrapulmonary type. Adult patients with scrotal paragonimus are rarely seen clinically and not widely reported in the literature. Here, we report 2 cases of scrotal paragonimiasis in adults and their treatment process.

Patient concerns: Two young males sought medical advice because of scrotal masses. Both patients had the previous history of eating uncooked river crabs and presented with palpable quasicircular nodules of about 1.5 × 1.0 cm in testicles, which were well-defined, resilient in tenderness. The bilateral inguinal lymph nodes were not enlarged.

Diagnosis and interventions: The 2 patients underwent scrotal mass resection; postoperative pathology examination confirmed scrotum paragonimiasis. Both the patients were administered praziquantel after operation.

Outcomes: They were followed up for 1 year and 4 years, respectively; both recovered well, free from recurrence. Scrotum color Doppler ultrasound examination found no obvious abnormality.

Lessons: Adult patients with scrotum paragonimiasis are rarely seen clinically. Moreover, its clinical manifestations are not typical that leads to missed diagnosis and misdiagnosis. Diagnosis of the disease needs to combine with disease histories, manifestations, and relevant auxiliary examinations. But the diagnosis can be confirmed only by histopathological examination. The main method for treatment of scrotal paragonimiasis is antiparasitic treatment. However, health education is crucial to prevent this disease and relapse.

Abbreviation: PPD = purified protein derivative.

Keywords: adult, diagnosis, prevention, scrotum paragonimiasis, treatment

1. Introduction

Paragonimiasis is a chronic parasitic disease caused by *Paragonimus westermani* and *Paragonimus skrjabini*. *P. westermani* induced diseases are mainly intrapulmonary types, while the extrapulmonary types involve brain, spinal cord, abdominal cavity, subcutaneous tissues, etc, and induce different symptoms. In contrast, *P. skrjabini* induced diseases are dominated by extrapulmonary types, and manifest as migration of adolesearia

and larvae in the body, which induces subcutaneous wandering masses and larva migrans. Scrotal paragonimiasis is mainly caused by *P. skrjabini*,^[1,2] and has been rarely investigated. Pulmonary and cerebral paragonimiasis, caused by *P. westermanii*, has been more commonly reported. Here, we reported 2 patients with scrotal paragonimiasis who were admitted to our hospital between 2012 and 2015.

2. Case reports

This study was approved by the Ethic Committee of the Affiliated Hospital of Zunyi Medical College.

Case 1, the patient was a 27-year-old man with a right scrotal mass. Three months ago, the patient accidentally found a bean-size nodule on the right side below the navel, which gradually migrated to the right scrotum. Case 2, the patient was a 21-year-old man with a left scrotal mass. Two years ago, the patient accidentally found a bean-size mass at the left scrotum, which progressively enlarged. There were no associated signs and symptoms such as fever and aversion to cold, cough and night sweat, frequent micturition and urgent urination, hematuria and dysuria, scrotal pendent swells, pruritus. They showed normal health, diet, sleep, urination, and defecation. They denied histories of hepatitis, tuberculosis, and other infectious diseases. Physical examination showed absence of abnormalities in the heart, lung, abdomen, extremities, and bilateral testes. Physical examination in case 1 and case 2 revealed quasi-circular palpable nodule of about 1.5 cm × 1.0 cm in size in the posterior part of the right epididymis and in the head of the left epididymis

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TL and YD contributed equally to this work.

All procedures performed in this study were in accordance with the ethical standards of the institution, the hospital affiliated to Zunyi Medical College. Informed consent was obtained from the patient included in this study.

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Urological Department, the Affiliated Hospital of Zunyi Medical College, Zunyi, China.

* Correspondence: Guobiao Liang, Urological Department, the Affiliated Hospital of Zunyi Medical College, Zunyi 563000, China (e-mail: lgb1111@126.com).

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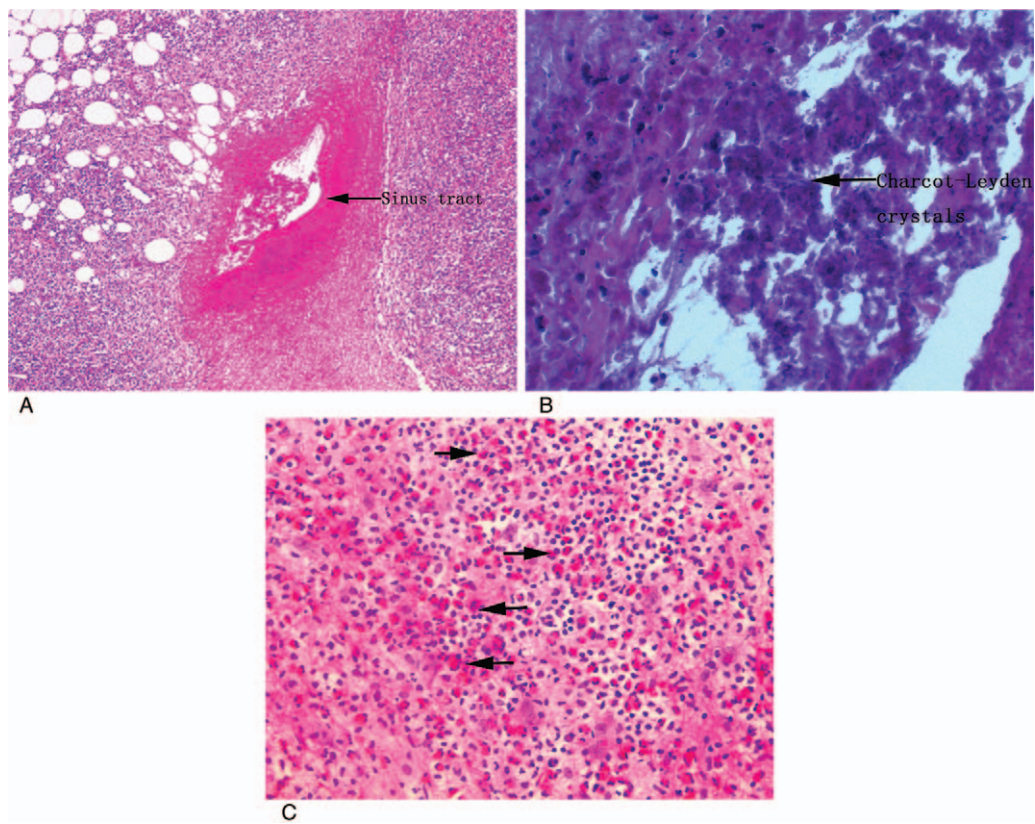


Figure 1. Case 1: Pathological results. (A) Sinus tract (HE, $\times 100$). The arrow indicates the sinus tract. (B) Charcot-Leyden crystals are visible in the sinus tract (HE, $\times 400$). The arrow indicates the Charcot-Leyden crystals. (C) Eosinophil infiltration (HE, $\times 400$). The arrow indicates the eosinophil granulocyte.

respectively. The nodules were mobile, well defined with resilient texture and smooth surface. Auxiliary examination showed normal blood routine, erythrocyte sedimentation rate, liver and kidney functions, urine and stool test, as well as chest radiography, negative PPD (purified protein derivative) test, and acid-fast stains on urinary sediment. The results of color doppler ultrasound of case 1 and case 2 are change of ultrasonography of tissue below cauda epididymidis, and disordered, inhomogeneous echo from the left spermatic cord running area, possibly inflammatory. The 2 patients are diagnosed of right scrotal nodules and left scrotal nodules, respectively. All relevant examinations were performed to exclude surgical contraindications. Both the patients underwent excision of scrotal mass under epidural anesthesia. During the surgery, the mass was found to be located between the dartos layer and tunica vaginalis of the scrotum and on top of the left testis respectively, firmly adhered to the spermatic cord and testis tunica vaginalis, with a size of $1.5\text{ cm} \times 1.0\text{ cm}$, gray and incompletely capsuled; white sticky discharge was visible during the surgery. The mass was sent for pathologic examination and the histopathology confirmed a diagnosis of paragonimiasis. The histopathological feature was more apparent in case 1 with numerous eosinophilic infiltration, topical sinus tract, and Charcot-Leyden crystals in the sinus tract (Fig. 1). The main histopathological feature of case 2 showed formation of sinus with infiltration of numerous eosinophils, lymphocytes, plasmacytes and mononuclear macrophages, numerous multinuclear giant cell proliferation, and eosinophilic abscesses in the mass as well, which was surrounded with proliferated epithelioid cells and a few multinuclear giant cells (Fig. 2). No parasitic ovum and

polypide was visible in both cases. According to the treatment criteria for paragonimiasis, the patient was postoperatively given praziquantel for a course of treatment. The dose was 25 mg/kg , 3 times a day for continuous 3 days. They were followed up for 4 years and 1 year, respectively. Scrotal ultrasound did not show abnormalities or lymph node enlargement in inguinal and other regions. It was later revealed by the patients that they have a previous history of eating uncooked river crabs.

3. Discussion

Paragonimiasis is a natural focal disease, which is caused by *P. westermani*, *P. skrjabini*, and other paragonimuses in the human body. It belongs to zoonotic helminthiasis. The main sources of infection include felines, canines, weasels, and other animals. The spread of the disease requires alternate hosts, the major one being snails followed by crabs and crawfishes. Patients become sick after drinking nonboiled water containing encysted cercariae, or eating uncooked, half-cooked, pickled, or liquor-saturated crabs or crawfishes.^[3] This disease is widely prevalent, and mainly distributed in Asia. In China, paragonimus is found in 24 provinces and cities, of which Zhejiang province and Northeast areas mainly have *P. westermani*, while Sichuan, Guizhou, Hunan, Hubei, Jiangxi, Henan, and Shanxi mainly have *P. skrjabini*.^[4,5]

Clinically, scrotal paragonimiasis is very rare, and mainly found in children. It is caused by *Paragonimus adolescentis* passing through the tissues of the human body, and characterized by the formation of mutually connected sinus tracts and multilocular small cysts. The pathogenesis of paragonimiasis is

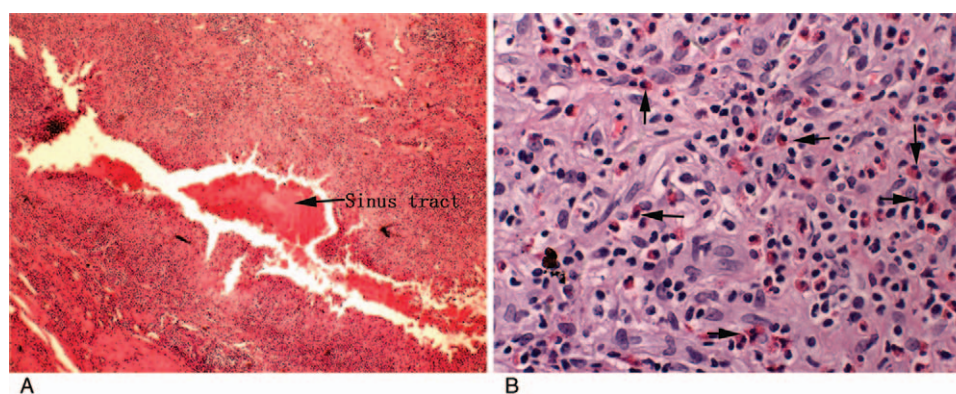


Figure 2. Case 2: Pathological results. (A) Sinus tract (HE, $\times 40$). The arrow indicates the sinus tract. (B) Eosinophil infiltration (HE, $\times 400$). The arrow indicates the eosinophil granulocyte.

as follows.^[6] First, the polypides migrate into the tissues and cause mechanical damage to tissues or organs. Second, metabolites of polypides, or decomposition products of dead polypides or dead ovum as foreign proteins can induce immunological reaction in the human body. Third, ovum can also induce foreign body granulomas in some regions. Typical pathological manifestations under microscopy include large areas of necrotic tissues and formation of sinus tracts, which are surrounded by massive eosinophil infiltration, formation of Charcot–Leyden crystals and fibroblastic proliferation, while occasionally ovum or polypides can be found in the lesion.

In the human body infected with *P. skrjabini*, polypides are generally immature, and mainly manifest as larva migrans that can be classified into cutaneous and visceral types.^[3,7] Scrotal paragonimiasis belongs to the cutaneous type. This type of larva migrans mainly appears as wandering subcutaneous masses or nodules of different sizes, normally ranging from 1 to 3 cm, but some can be of the size of an egg. Also, there may be single or multiple masses, which are spherical or elongated, with unclear boundary and normal skin surface. Sometimes, there are palpable streak fiber blocks between masses. The masses are often found in the chest, abdomen, and lumbodorsal part, but can also be found in the head and neck, fossa axillaris, extremities, hip, inguina, scrotum, etc.^[8,9]

In this study, both patients visited the hospital due to scrotal nodules, of which case 1 showed typical wandering subcutaneous nodules. However, because both patients were adults and were admitted without detailed disease histories, paragonimiasis was not considered at admission. Scrotal paragonimiasis has a low diagnosis rate, and is prone to misdiagnosis and missed diagnosis. The reasons may be as follows. First, with the improvement of living standard as well as medical and health conditions, the incidence of parasitic diseases is decreasing. Furthermore, scrotal paragonimiasis mostly occurs in children, and is rarely found in adults. Second, clinicians lack knowledge of this disease, and do not comprehensively inquire about disease histories. Third, some patients do not exhibit typical manifestations, such as wandering subcutaneous masses or nodules. Fourth, most masses do not induce pain, and patients are often misdiagnosed as tuberculosis of scrotum if they are living in areas with high incidence of tuberculosis. Both patients in this study were not preoperatively diagnosed with this disease, but diagnosed by postoperative pathology. After the surgery, disease histories were inquired again, which indicated that both patients had eaten uncooked river crabs before 6 months to 1 year. Therefore, clinicians should be vigilant

of scrotal paragonimiasis. In case of unexplained scrotal masses, scrotal paragonimiasis should be highly suspected under the following conditions: First, patients living in remote mountainous or paragonimiasis prevalence areas; second, patients with histories of consuming uncooked or half-cooked crabs or crawfishes; third, patients with increased eosinophils for unknown reasons; and fourth, patients with wandering or nonwandering (accompanied with 1 of the above 3 items) scrotal masses.

Early diagnosis, treatment, and health education are very important for the prevention and treatment of this disease. For suspected cases of paragonimiasis, a detailed history including their recent diet has to be enquired and relevant examinations line eosinophil count and paragonimus antigen dermal sensitivity test must be conducted. For suspicious subcutaneous nodules, needle aspiration biopsy and smear cytology must be performed to detect polypides, ovum, Charcot–Leyden crystals, and eosinophils. But in clinical practice, surgical excision is usually performed, as this procedure not only helps in diagnosis but also achieves the aim of treating disease.^[3] Scrotal paragonimiasis can be successfully treated by drugs; thus, surgery is usually unnecessary. The drug of choice is praziquantel 25 mg/kg, 3 times a day for 3 days.^[10] This disease can be prevented by strengthening health education. For example, nonboiled stream water, uncooked or half-cooked river crabs or crawfishes, and pickled or liquor-saturated crabs should not be consumed. Active treatment should be provided to patients with suspected paragonimiasis.^[11]

In summary, scrotal paragonimiasis is a rare chronic parasitic disease, which mainly occurs in children, but rarely in adults. It has atypical manifestations and is prone to missed diagnosis and misdiagnosis. Diagnosis of the disease should involve disease histories, manifestations, and relevant auxiliary examinations. Confirmed diagnosis requires pathological examinations (eggs found in biopsy). The main therapy for scrotal paragonimiasis is antiparasitic treatment. Some patients may have an obvious scrotal mass, but other reasons for the mass such as tumors cannot be excluded. Therefore, mass excision combined with antiparasitic treatment is the only means for diagnosis and radical treatment of this disease.

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Author contributions

Conceptualization: A. Chen, H. Li, J. Du, L. Yu, X. Wang, Y. Du, Y. Wang, Z. Chen.

Writing – original draft: T. Liang.

Writing – review & editing: G. Liang.

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