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Case report

A case report of human primary renal cystic echinococcosis

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ABSTRACT

In humans, solitary renal involvement or primary renal echinococcosis is rare, accounting for about 2–4 % of cases. Usually, patients shpw no obvious symptoms, but they can manifest as renal pain, renal mass, gross hematuria, and hydatiduria in rare cases. We report a case of primary renal cystic echinococcosis, which was originally misdiagnosed as a tuberculous renal abscess.

Introduction

Echinococcosis is a zoonotic parasitic disease caused by the Echinococcus spp., which seriously endangers human health and animal husbandry development [1]. Echinococcosis, distributed worldwide and spread with the development of animal husbandry, has become an important global public health and socio-economic problem [2]. The growth rate of echinococcosis reportedly increased significantly from 2004 to 2013 [3], although its incidence has been decreasing in some countries through the implementation of effective control programs [4]. In China, Qinghai is a severe endemic area of echinococcosis. The liver is the most frequently involved site of echinococcosis, followed by the lung. In this paper, we report a case of primary renal cystic echinococcosis, who was diagnosed accurately before operation due to previous tuberculosis and tuberculous epididymitis.

Case record

The publication of the patient's data in this report was approved by the Ethical Committees of the Qinghai University Affiliated Hospital (approval number: AF-RHEC-0103-72), and written informed consent was obtained from the patient.

A 34-year-old male patient was admitted for urinary surgery to treat right scrotum swelling and pain lasting 10 days. He had lived in the urban area of Xining City, Qinghai Province, since he was a child, and had no contact history with cattle, sheep, dogs, or other animals. He had a history of pulmonary *Mycobacterium tuberculosis* infection 8 years ago and recovered after anti-tuberculosis treatment. A physical examination showed that the right scrotum was enlarged to about 3.0 × 2.0 cm; testicular palpation texture was hard with squeezing pain; a 0.2 cm scrotal skin rupture was seen below the mass, and yellow purulent fluid was seen to flow out; a fistula tract was not formed, and it was not drained. The patient's temperature was 36.8 ℃. Color Doppler ultrasound of the genital organs showed the enlargement of the right epididymis. A chest computed tomography (CT) imaging scan showed that there were no sequelae of tuberculosis or other lesions (Fig. 1). A further abdominal CT scan showed circular cystic lesions in the left kidney, with the largest cystic lesion being about 6.6 × 5.6 cm (Fig. 2A). A magnetic resonance imaging (MRI) scan showed long T1 and long T2 signals in the cystic lesion; tortuous structures were found inside the largest lesion, and cystic wall enhancement was observed on an enhanced scan (Fig. 2B). CT angiography (CTA) showed compression of the left renal artery and left renal vein (Fig. 3). Renal dynamic imaging showed that the glomerular filtration rates of the kidneys were 35.5 mL/min (left) and 59.2 mL/min (right) (Fig. 4). A fine-needle aspiration testicular biopsy showed the presence of some necrotizing granulomas. The biopsy samples of the testicular lesions were found to be positive for *Mycobacterium tuberculosis* using PCR. Laboratory tests showed neutrophils of 78 % (normal range, 40–75 %), lymphocytes of 14.1 % (normal range, 20–50 %), C-reactive protein of 20.4 ng/L (normal range, 0–6 ng/L), alkaline phosphatase of 184 U/L (normal range, 45–125 U/L), and γ-glutamyltransferase of 100 U/L (normal range, 10–60 U/L). The γ-interferon release test was positive. The human echinococcosis IgG test was negative. The urine tuberculosis culture was negative. According to the laboratory examination, imaging examination, pathological examination and previous tuberculosis infection history of the patient, the

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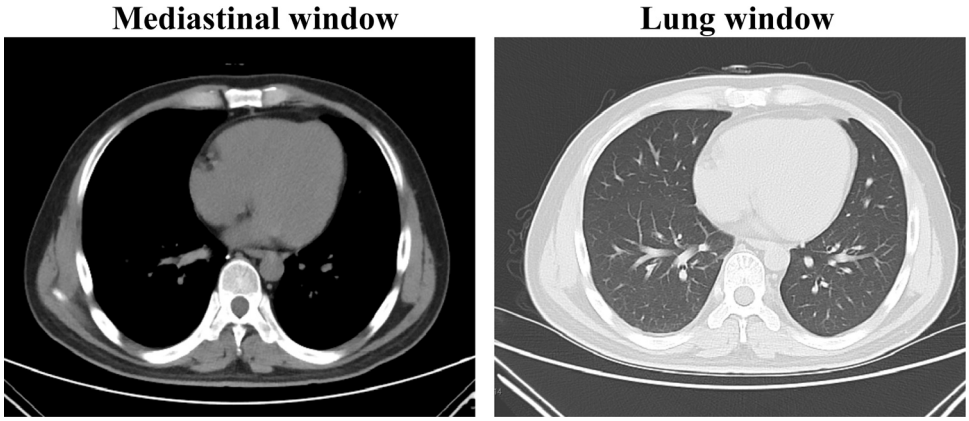

Fig. 1. Chest computed tomography image.

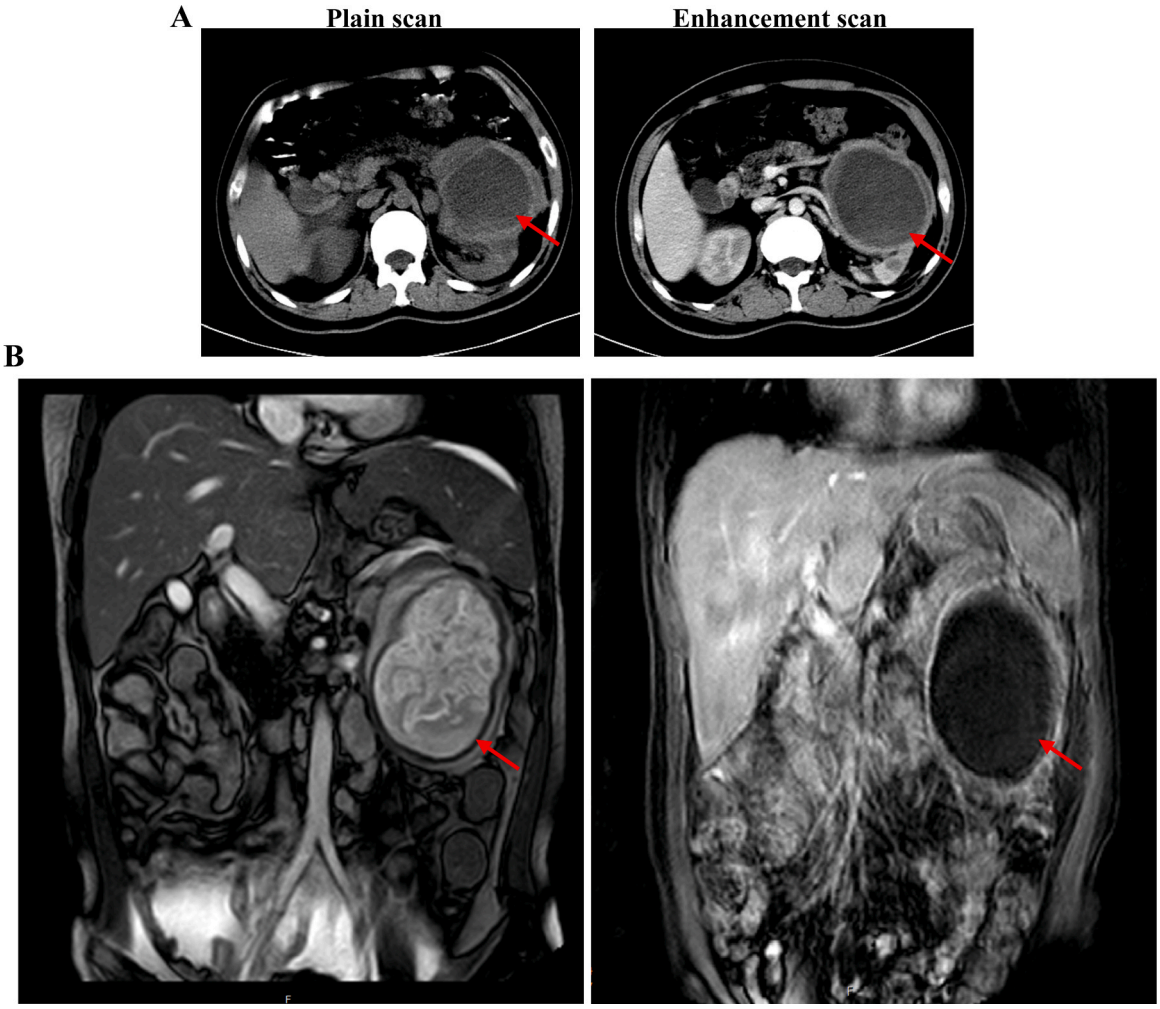

Fig. 2. Preoperative abdominal computed tomography and magnetic resonance imaging. (A) Computed tomography showed a large space-occupying lesion in the left kidney. The largest cystic lesion was approximately 6.6 × 5.6 cm in size (red arrow). (B) A magnetic resonance imaging scan showed multiple space-occupying lesions in the left kidney. The cystic lesion (red arrows) in the kidney showed long T1 and long T2 signals, and the cystic wall was slightly enhanced on an enhanced scan.

patient was diagnosed as having right epididymal tuberculosis and a left renal tuberculous abscess. The patient began to take isoniazid, rifampicin and ethambutol (once a day, 300 mg each) orally for anti-tuberculosis treatment.

The patient first underwent resection of the right epididymal lesion. A postoperative pathological examination showed coagulation necrosis, Langerhans giant cells and epithelioid cells in the lesion. One month later, he underwent kidney lesion resection under general anesthesia. During the operation, obvious adhesion was found with the spleen, omentum, upper pole of the left kidney, and renal hilus. Because it was

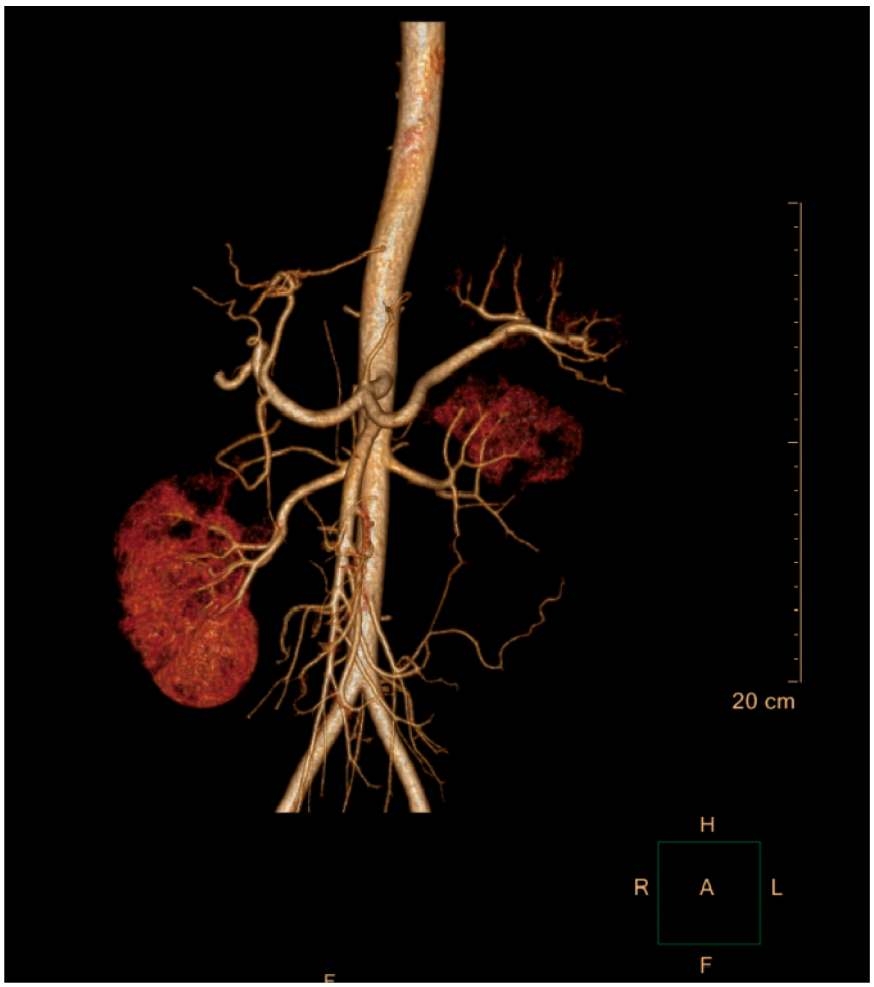

Fig. 3. CT angiography showed renal perfusion.

difficult to separate the adhesion between the upper pole of the left kidney and the lesion, the upper part of the left kidney was resected. There were multiple fluid-filled cystic lesions inside the resected lesion (Fig. 5A). A pathological examination of a larger cyst showed the presence of protoscoleces, germinal cells, and a PAS-positive laminated layer, but no typical tuberculous pathological changes were observed (Fig. 5B,C). After the operation, the lesion polymerase chain reaction (PCR) test was negative for mycobacterium tuberculosis and positive for *Echinococcus granulosus* (Fig. 6). A pathological examination confirmed that the renal lesions of the patient were cystic echinococcosis. The patient recovered well after the operation without surgical complications. The patient took oral albendazole (400 mg twice a day) for at least two years after the operation. During the 6-month follow-up, an ultrasound examination showed that there was no active infection or new lesions at the surgical site. The patient was followed up one year later, and no echinococcosis or tuberculosis had occurred in other organs.

Discussion

The life cycle of Echinococcus involves two mammalian hosts; usually, the final host are carnivores, and the intermediate hosts are herbivores [5]. Human beings are accidental intermediate hosts and are mainly infected by eating food or drinking water contaminated by eggs or through direct contact with infected soil or dogs. The increased risk of Echinococcus infection in the definitive host (dogs and foxes) is related to internal organ feeding, a lack of anti-helminth drug treatment, and insufficient health education for human beings [6]. When Echinococcus eggs are ingested by intermediate hosts (such as humans, bovines, and sheep), embryos break out of the eggs and enter the systemic circulation through the intestinal mucosa [7]. Larvae can transfer through blood circulation and lymph circulation. The liver is the most frequently affected site of echinococcosis, followed by the lung. Through the blocking effect of the liver and lung, the incidence of echinococcosis in other organs is relatively low.

There are no obvious clinical symptoms in the incubation period of echinococcosis, and most patients find lesions by chance, as in the case reported here. Compression or rupture of a cyst in the liver can result in pain in the right upper abdomen, cholangitis, and an allergic reaction [8]. The growth rate of cysts is relatively slow, with an incubation period that lasts about 5–15 years. Usually, the diameter of cysts reaches about 10 cm before showing clinical symptoms [8]. However, the clinical manifestations of extrahepatic primary echinococcosis are mostly atypical. For this patient, no clinical symptoms were shown until the time of treatment. Fortunately, we found the lesion when screening for urinary tuberculosis, but we made a wrong diagnosis before the operation. The discovery of liver echinococcosis lesions has important guiding significance for the diagnosis of extrahepatic echinococcosis. The involvement of renal echinococcosis is relatively rare, accounting for only about 2–4 % of echinococcosis [9]. In this case, no liver or lung lesions were found during imaging examinations, which made the diagnosis of echinococcosis relatively difficult. Cystic/solid renal lesions were not determined to be atypical in the imaging examination because of their small size. Because the patient had a *Mycobacterium tuberculosis* infection in the past, both the pathological biopsy and PCR examination of the testicular lesions were positive, which interfered with the accurate diagnosis of renal echinococcosis.

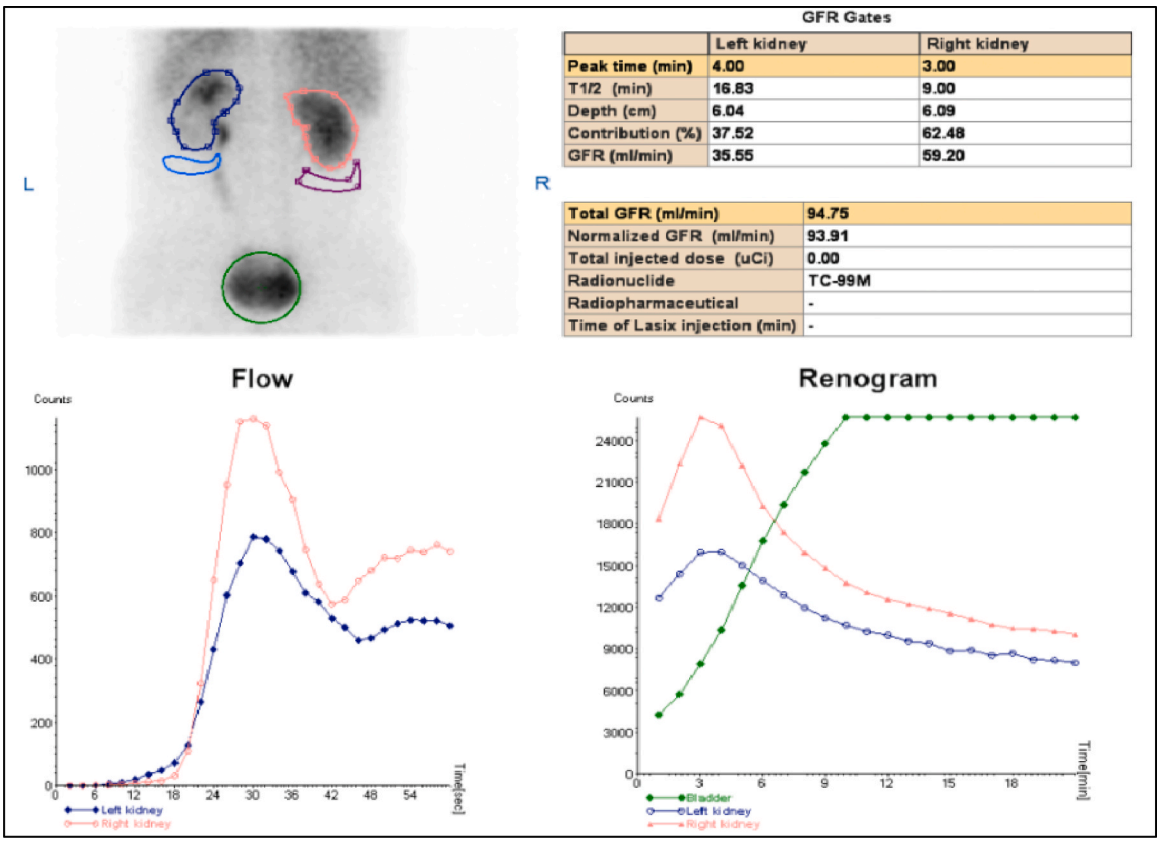

Fig. 4. Renal dynamic imaging showed impaired left renal function.

The diagnosis of echinococcosis depends on life history and serological and imaging examinations. Most patients infected with echinococcosis come from pastoral areas and have a clear history of close contact with cattle, sheep, dogs, and other animals. The patient had lived in the city for a long time, had no life history in pastoral areas or epidemic areas, and had no habit of eating raw vegetables or meat. The infection route is still worth tracing. Serological detection methods for echinococcosis include indirect hemagglutination (IHA), indirect fluorescent antibody (IFA), an enzyme-linked immunosorbent assay (ELISA), and latex agglutination. These detection methods can detect antibodies against crude or purified hydatid cyst fluid antigens [10]. It has been reported that the sensitivity of these detection methods to liver echinococcosis is 85–98 %, but the sensitivity to lung echinococcosis is obviously lower [11]. In addition, the sensitivity of detection is affected by cyst maturity, cyst wall integrity and lesion infiltration. After cyst wall rupture, the detection rate is significantly increased when the cyst fluid outflows and a host immune reaction occurs [11]. However, these detection methods also have cross-reactions with other parasitic diseases (such as cysticercosis) and non-parasitic diseases (such as malignant tumors and liver cirrhosis) [10]. In this case, the serological IgG test for echinococcosis was negative after the operation, which may be directly related to the fact the cyst wall was intact. However, it is difficult to diagnose echinococcosis when the serological IgG of echinococcosis is negative. However, fortunately, we completely removed the lesion without rupture during the operation, which is very important to prevent the recurrence of postoperative diseases.

Radical surgical resection combined with chemotherapy is the first choice for the treatment of echinococcosis. For a single large cyst, puncture, aspiration, injection and relapse therapy (PAIR) can also be used [12], whereby the parasites in the cyst cavity are killed by injecting alcohol or hypertonic saline into the cyst [12]. The diagnosis and treatment of patients with a mixed infection of two types of echinococcosis are more complicated. The surgical treatment of patients with a mixed infection should be based on ensuring the safety of the operation, and all lesions should be removed by radical surgery as much as possible. The cystic lesions should be completely removed during the operation to prevent cystic fluid leakage. According to the recommendations of the World Health Organization, for patients with echinococcosis, drug treatment (such as albendazole) should be started 1 month before surgery to kill most surviving parasites [12].

Conclusions

Missed diagnosis, misdiagnosis or improper treatment will result in the recurrence and multiple metastases of echinococcosis. Although we did not make an accurate diagnosis before the operation, we completely removed the lesion and achieved relatively satisfactory results. For patients with previous infectious diseases, a differential diagnosis is particularly important. Finally, all patients with echinococcosis infection undergoing surgical treatment should be treated with the oral administration of benzimidazole drugs according to the diagnosis and treatment standards of echinococcosis after operation.

Ethical approval

We declare that the approval of the ethics committee is not necessary.

Consent

We declare that the consent is not necessary.

Author agreement

We agree to the eventual publication of the article if it is accepted.

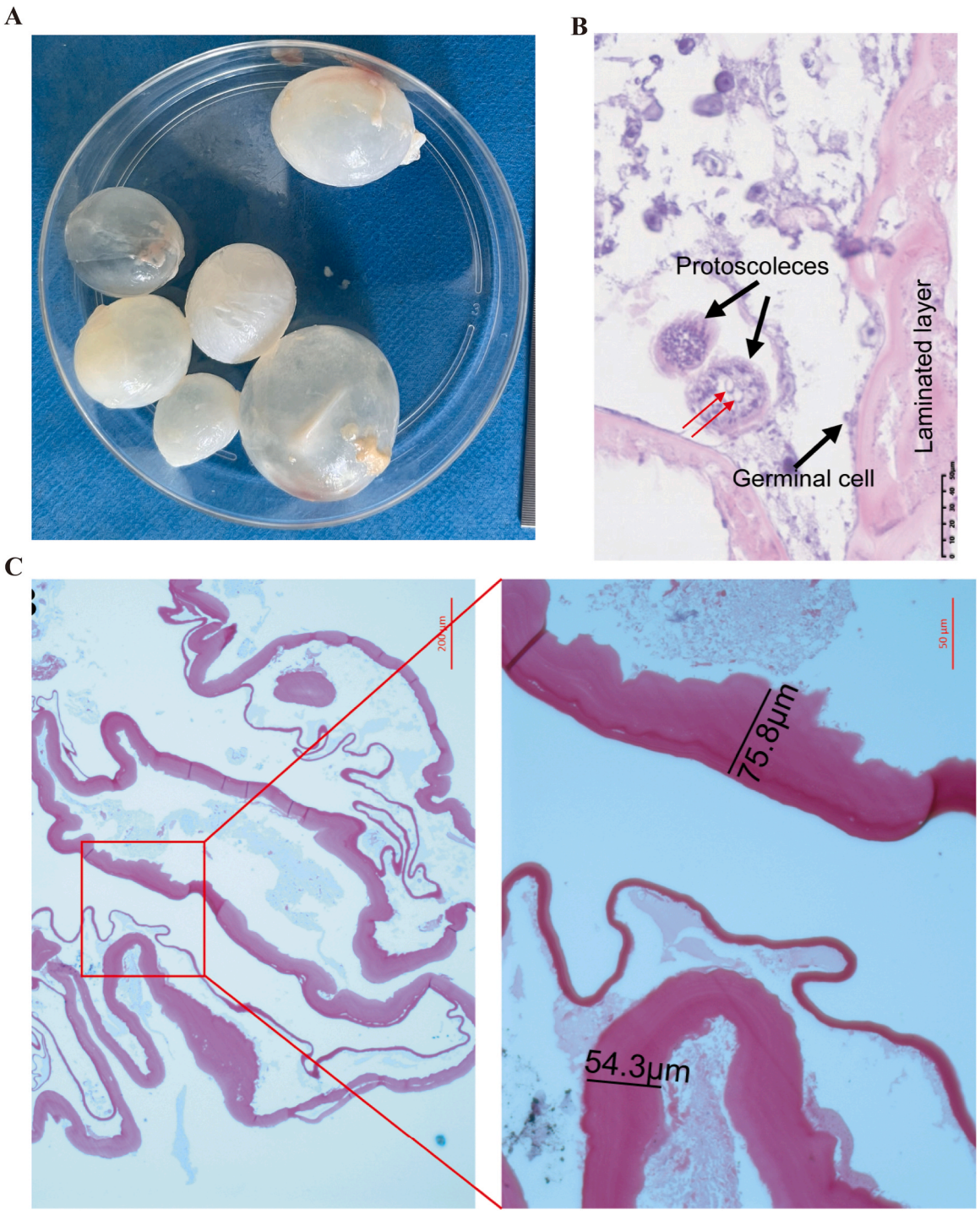

Fig. 5. Pathological examination of the excised lesion. (A) Partial lesion gross morphology. (B) Hematoxylin and eosin staining of cystic lesion. Protoscoleces were observed in the cyst. (C) A thicker laminated layer was observed in the cyst. Periodic acid–Schiff (PAS) stain presented a strongly PAS-positive basophilic laminated layer (red arrows).

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Author statement

We declare that we have read the reviewers' remarks and have taken them into account.

CRediT authorship contribution statement

Zhuoma Dawa: Writing – original draft. **Chuanchuan Liu:** Writing – review & editing, Conceptualization. **Haining Fan:** Writing – review & editing, Conceptualization.

Declaration of Competing Interest

We declare that we have no conflict of interest.

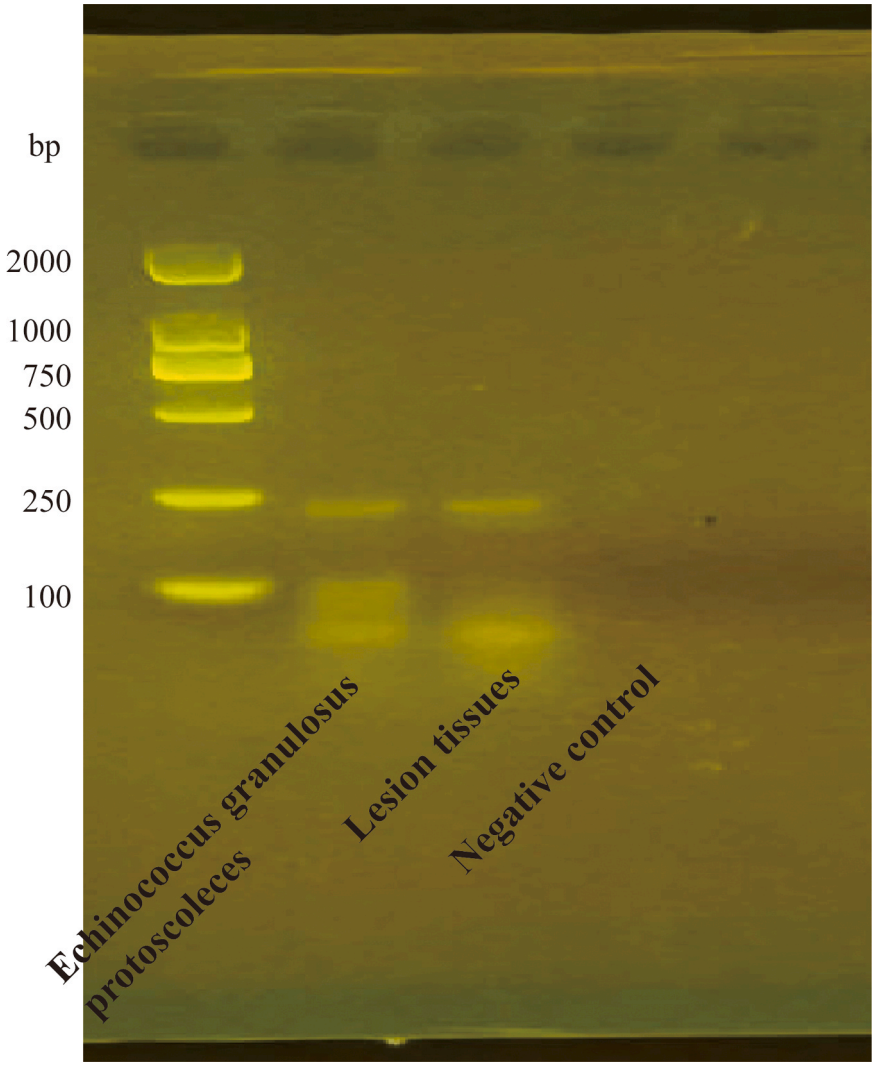

Fig. 6. Identification of *Echinococcus granulosus* nad1 genes via agarose gel electrophoresis.

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