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Case Report

Disseminated cysticercosis incidentally diagnosed in a patient with distal cholangiocarcinoma: A case report ☆,☆☆

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ABSTRACT

Cysticercosis, a major health issue in developing countries, is caused by the larval stage of *Taenia solium*. Disseminated cysticercosis (DCC), which is characterized by widespread cysticerci in various tissues, is rare and often asymptomatic. Here, we report the case of a 50-year-old man from rural Nepal with distal cholangiocarcinoma and DCC involving the skin, brain, orbit, tongue, soft palate, heart, and abdominal organs. Despite the presence of abdominal pain, obstructive jaundice, anemia, and significant weight loss—symptoms indicative of biliary malignancy—there were no symptoms typical of DCC. Diagnostic imaging confirmed DCC and stomach-preserving pancreaticoduodenectomy was performed. Histopathological examination of the periampullary mass revealed distal cholangiocarcinoma. Postsurgical treatment for DCC included steroids, carbamazepine, and antiparasitic therapy with albendazole. The coexistence of cysticercosis and neoplasia, though uncommon, necessitates thorough diagnostic evaluation. This case underscores the clinical complexity and highlights the need for comprehensive management of concurrent conditions.

Introduction

Cysticercosis, a prevalent public health issue, particularly in developing nations, is primarily caused by *Cysticercus cellulosae*, the larval form of the tapeworm, *Taenia solium*. Disseminated cysticercosis (DCC) occurring as a result of the widespread invasion of the cysticerci via the intestine through entering hepatic circulation to various tissues is a rare manifestation of cysticercosis [1] and still rarer is asymptomatic

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presentation. Organs invaded include subcutaneous tissue, skeletal muscles, brain, lungs, eyes, liver, and occasionally the heart. Pseudomuscular hypertrophy, palpable subcutaneous nodules, seizures, and abnormal mentation characterize the syndrome of DCC [2].

We report a case of DCC involving the skin, brain, orbits, tongue, soft palate, heart, and various abdominal organs in a patient with distal cholangiocarcinoma.

Case presentation

A 50-year-old married man from rural Nepal was referred to a tertiary care center with complaints of generalized yellowish discoloration and abdominal pain persisting for 2 months, accompanied by generalized itching and significant weight loss (19% of body weight lost in the past 6 months). He denied having fever, abdominal distension, or vomiting. He had no history of blood transfusions, intravenous drug use, piercing, unsafe sexual contact, similar previous illnesses, or significant familial, medical, surgical, traumatic, or vaccination history.

During the examination, the patient appeared thin, with signs of pallor and icterus. No supraclavicular, cervical, or inguinal lymphadenopathies were observed. Multiple soft, mobile, nontender subcutaneous nodules were palpable diffusely over the anterior chest, upper back, neck, and both upper and lower limbs. An abdominal examination revealed a lump in the right upper quadrant, which moved with respiration. Neurological, cardiovascular, and chest examinations were unremarkable.

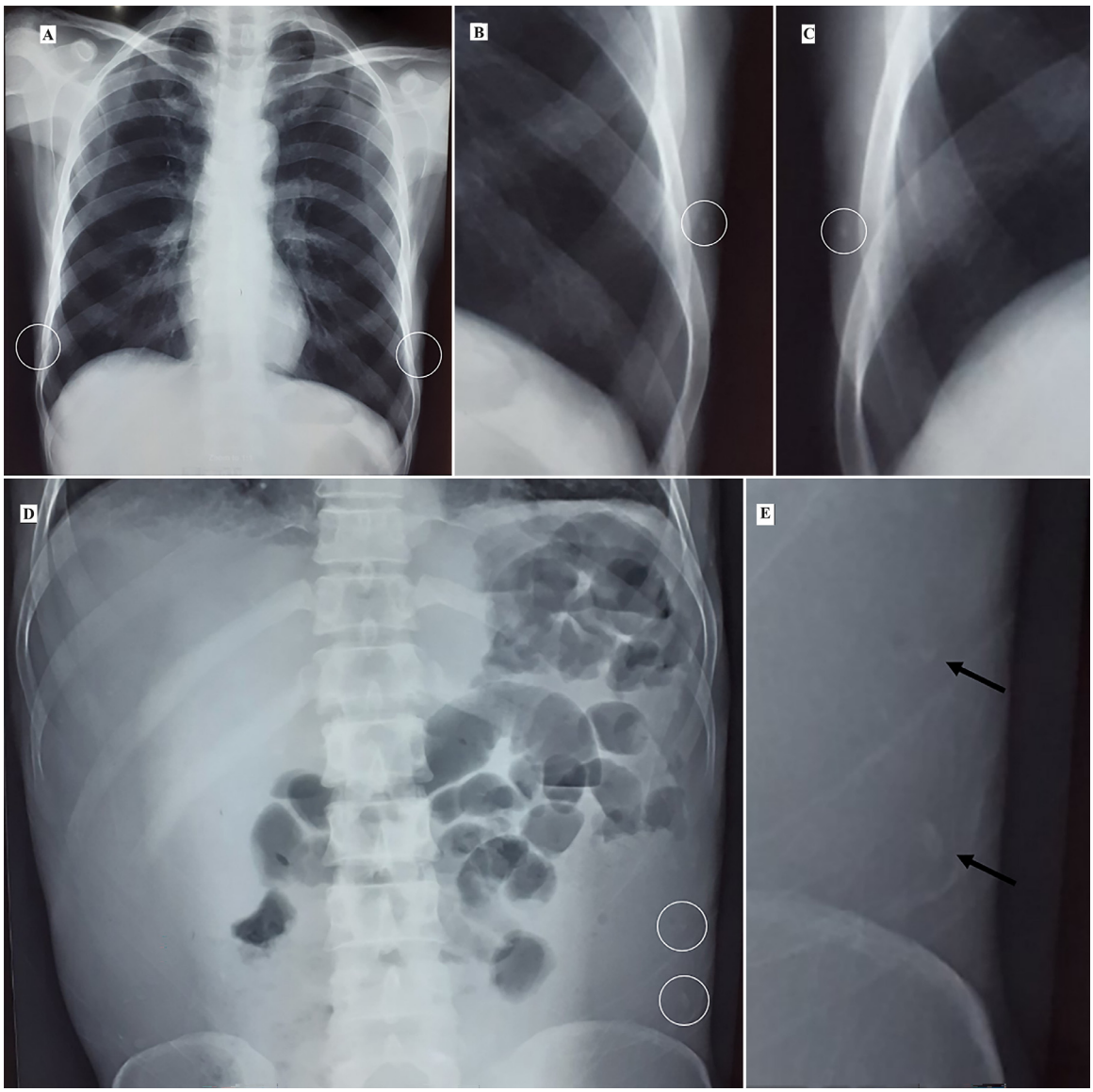

Fig. 1 – Plain radiograph showing small soft tissue calcification (circled areas and black arrows) giving typical "rice grain appearance". (A) In bilateral chest wall of chest radiograph. (B and C) Magnified view of the chest radiograph. (D) In left lateral abdominal wall of erect abdominal radiograph. (E) Magnified view of the abdominal radiograph.

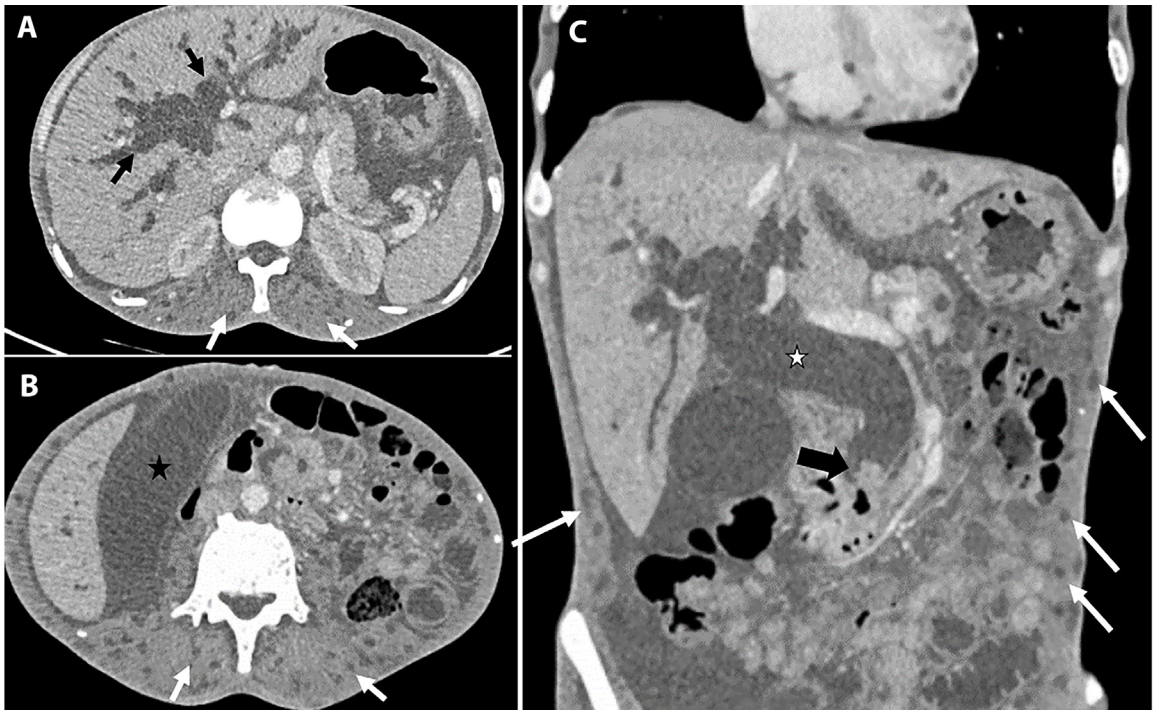

Fig. 2 – Contrast enhanced images: (A) Axial image showing severe dilatation of IHBD (black arrows). (B) Axial image showing distended Gall bladder (black star). (C) Coronal image showing distal CBD mass (black boxed arrow) with dilated common bile duct (white star). Multiple granulomas are seen in bilateral paraspinal muscles (white short arrows) and along the peritoneum (white long arrows).

Table 1 – Various blood parameters of the patient upon admission.

Parameters	Result	Normal reference
Complete blood count		
Hemoglobin (gm/dL)	5.4	14.0-18.0
TLC (cells/mm^3)	10100	4000-11,000
Platelets(cells/mm^3)	623000	150,000-400,000
Red Cells (cells/mm^3)	1.5 million	4.5-5.5 million
PCV (%)	16.6	40-54
MCV (fL)	110	82-92
MCH (pg)	36	26-34
MCHC (%)	32	32-36
PT (seconds)	13.0	11-16
INR	1.28	
Liver Function Test		
Total Bilirubin (mg/dL)	19.8	0.3-1.2
Direct Bilirubin (mg/dL)	10.3	<0.2
ALP (U/L)	822	30-120
GGT (U/L)	347	11-50
ALT (U/L)	130	<50
AST (U/L)	60	<50
Total protein (gm/dL)	5.5	6.3-8.3
Albumin (gm/dL)	2.4	3.5-5.5
Tumor Markers		
CEA (ng/dl)	1.74	0-4
CA-125 (U/ml)	195	<35
CA 19-9 (U/ml)	26759	<19

ALP, Alkaline phosphatase; ALT, Alanine aminotransferase; AST, Aspartate aminotransferase; CEA, Carcinoembryonic antigen; GGT, Gamma glutamyl transferase; INR, International Normalized Ration; MCH, Mean Corpuscular Hemoglobin; MCHC, Mean corpuscular hemoglobin concentration; MCV, Mean corpuscular volume; PCV, Packed cell volume; PT, Prothrombin time; TLC, Total leukocyte count.

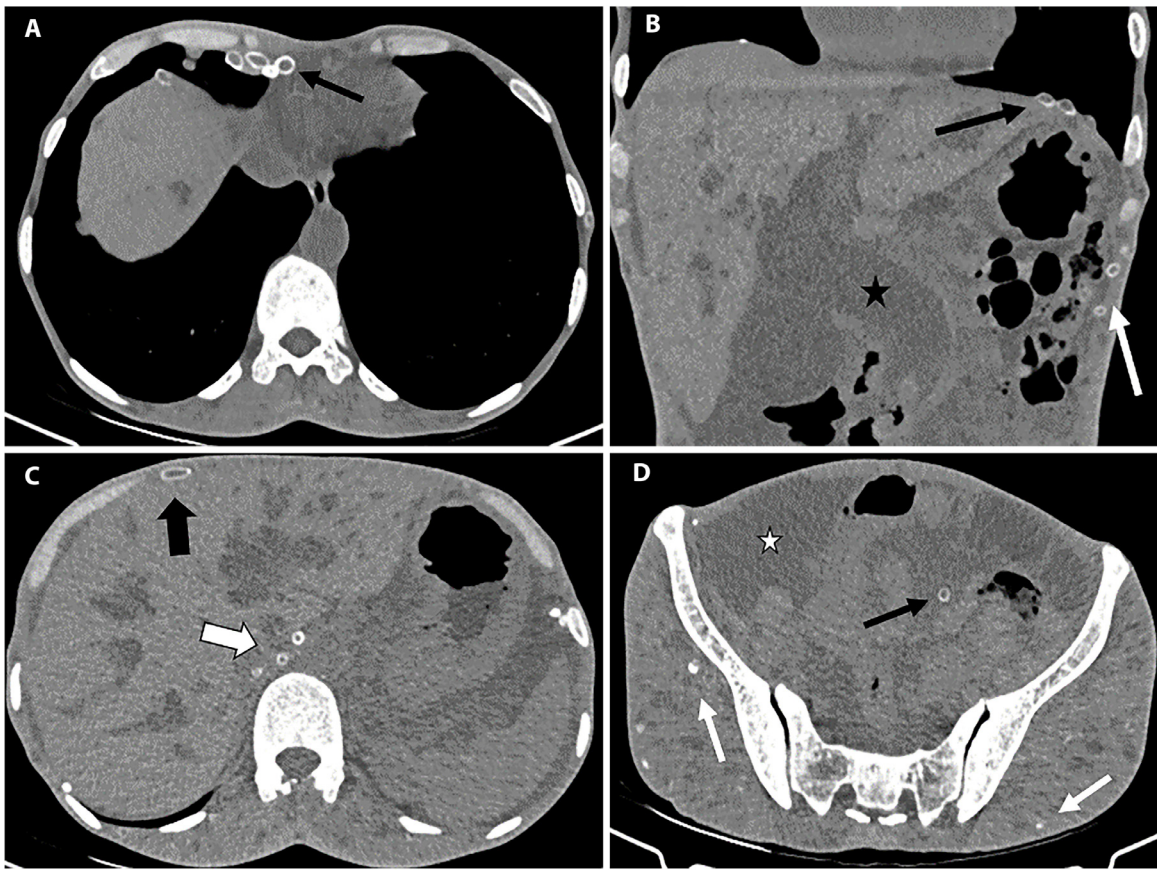

Fig. 3 – Noncontrast CT images of abdomen and pelvis showing multiple calcified lesions in various organs: (A) Along mediastinal pleura (black arrow). (B) Along left diaphragm (black arrow) and peritoneal lining (white arrow); dilated common bile duct also present (black star). (C) Along right crus of diaphragm (boxed white arrow) and liver capsule (boxed black arrow). (D) In mesentery (black arrow) and bilateral gluteal muscles (white arrow); ascites (white star) also present.

Blood investigations revealed severe anemia, thrombocytosis, obstructive jaundice, and elevated serum tumor markers including CA-125 and CA 19-9 (Table 1). The urinalysis and electrocardiogram (ECG) were normal. A plain radiograph of the chest and abdomen showed soft tissue calcification of the bilateral chest wall and abdominal wall (Fig. 1).

Abdominal and pelvic sonography revealed a markedly distended gallbladder (GB) with an approximately 2.4 × 1.2 cm isoechoic lesion in the distal common bile duct (CBD). This lesion caused abrupt luminal narrowing and resulted in upstream dilatation of the CBD and intrahepatic biliary ducts, suggesting a periampullary mass with obstructive biliopathy. To address this issue, a percutaneous transhepatic biliary drain was placed to facilitate bile drainage.

Furthermore, a contrast-enhanced computed tomography (CECT) scan of the abdomen and pelvis (Fig. 2) confirmed the sonographic findings revealing approximately 1.6 × 1.3 cm irregular soft tissue density in distal CBD, which showed heterogeneous enhancement on postcontrast images causing obstructive biliopathy. The scan also showed gallbladder distention (11.8 × 4.2 cm) with a dilated CBD measuring 3.2 cm and gross ascites. Additionally, multiple hypodense nonenhancing granulomas were observed in various locations including the pleura, peritoneal lining, diaphragm, abdominal wall muscles, liver capsule, mesentery, gluteal muscles, and within the myocardium and interventricular septum (Figs. 3-5). These findings suggest myo-cysticercosis and cardiac cysticercosis.

Moreover, CT images of the brain showed multiple cystic lesions with central calcification in the grey and white matter of bilateral cerebral hemispheres, right cerebellar hemisphere, pons, and subarachnoid spaces (Fig. 6). A contrast-enhanced magnetic resonance imaging (MRI) of the brain showed T1 multiple isointense lesions with central high signal intensity scolex in bilateral cerebral hemispheres, T2 weighted and FLAIR showing T2 high signal intensity cysts suppressed on FLAIR images with central scolex in bilateral cerebral hemispheres. MRI also revealed cysts in extraocular muscles, tongue, soft palate, and trapezius muscle (Figs. 7 and 8).

Western blot analysis and enzyme-linked immunosorbent assay for serum cysticercosis IgG antibody could not be done due to unavailability in our center. However, the presence of cystic lesions demonstrating the scolex, a pathognomonic feature of cysticercosis, helped us finalize the diagnosis.

An upper gastrointestinal endoscopy revealed circumferential ulcerative growth with slough at the D1-D2 junction.

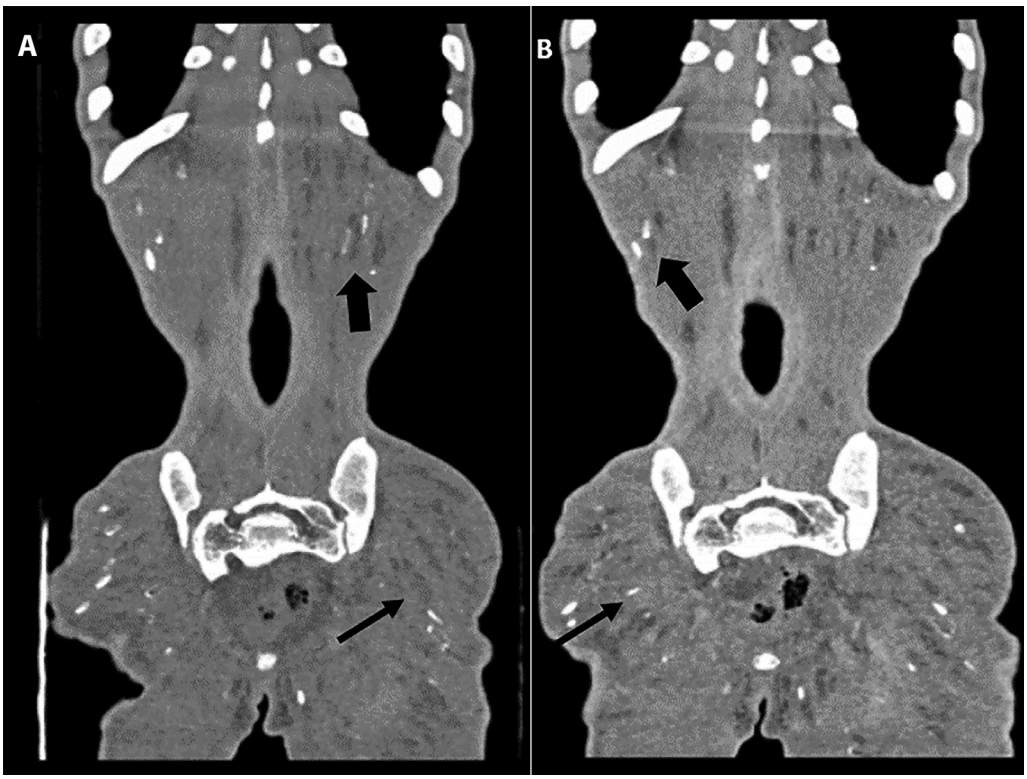

Fig. 4 – Coronal CT images showing multiple calcified and noncalcified granulomas in bilateral paraspinal muscles (black boxed arrows) and gluteal muscles (black arrows).

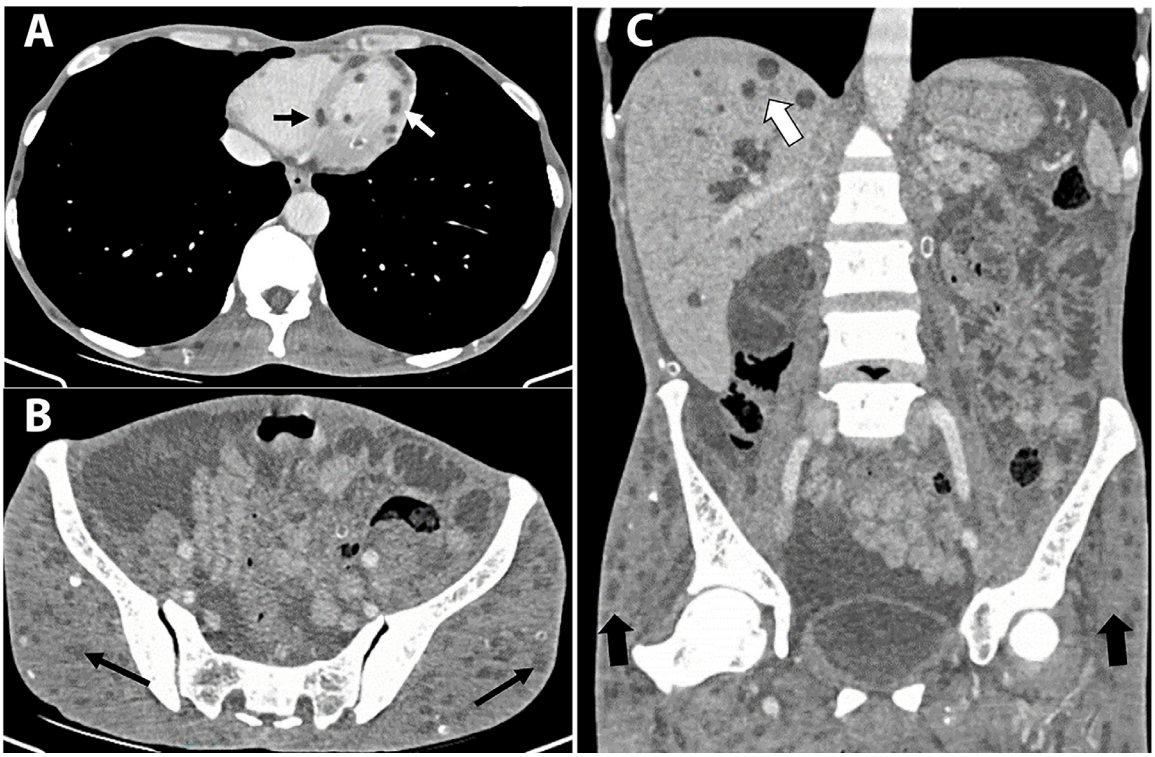

Fig. 5 – Post contrast CT images of abdomen and pelvis showing multiple granulomas in various organs: (A) In myocardium (short white arrow) and interventricular septum (short black arrow) of heart. (B) In bilateral gluteal muscles (black long arrow). (C) In liver (white boxed arrow) and muscles of pelvis (black boxed arrow).

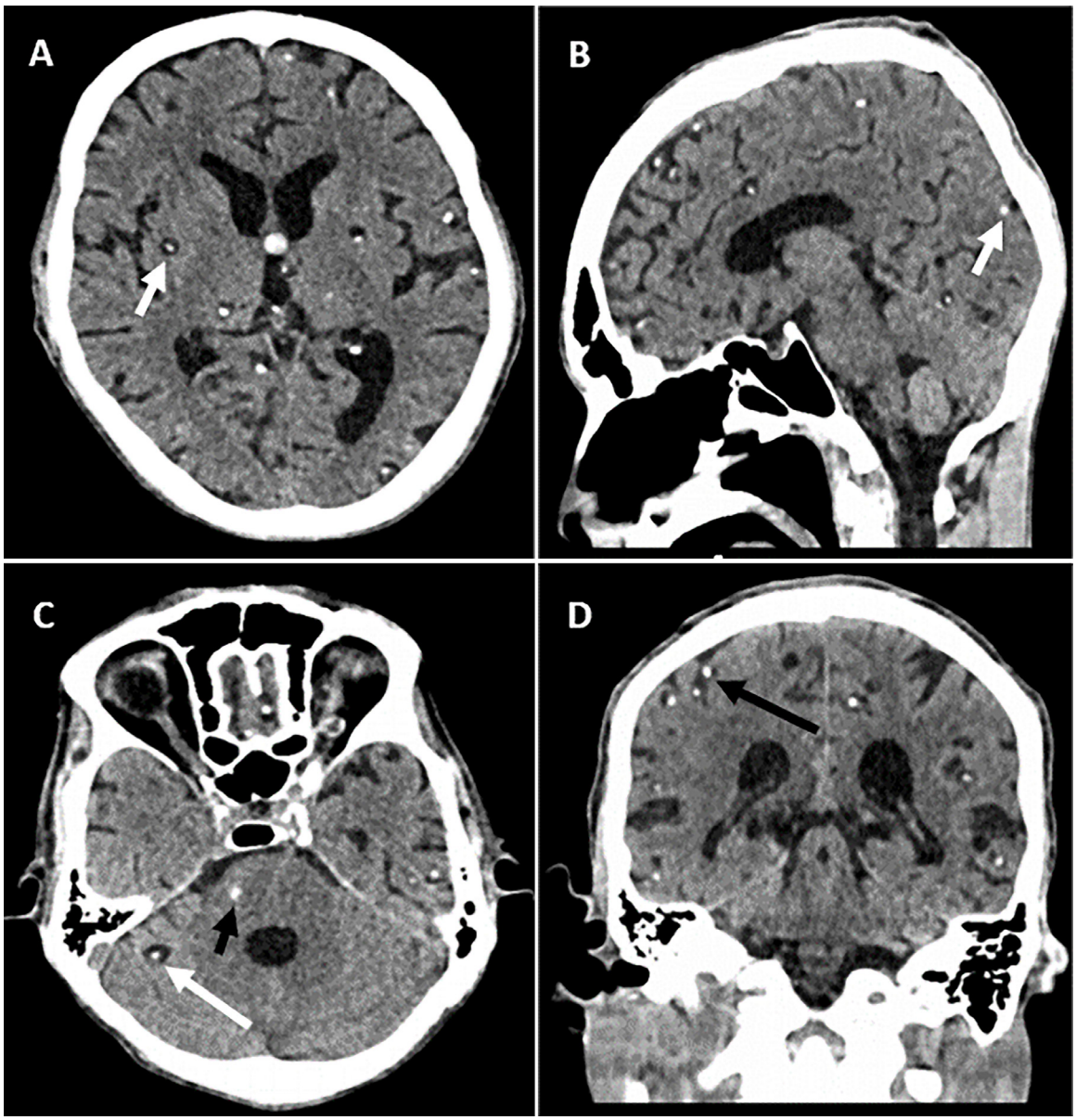

Fig. 6 – Noncontrast CT images of the brain showing multiple cystic lesions with central calcification in grey and white matter of bilateral cerebral hemispheres (short white arrows), right cerebellar hemisphere (long white arrow), pons (short black arrow) and subarachnoid spaces (long black arrow).

Biopsy from the ampulla showed ulcerative lesions with severe dysplasia while those from the D1, and D2 segments were negative for dysplasia. Ascitic fluid cytopathology was negative for malignant cells.

Based on these findings, a provisional diagnosis of periampullary carcinoma with disseminated cysticercosis was established. A stomach-preserving pancreaticoduodenectomy was performed, revealing a 3 × 3 cm hard mass in the distal CBD. Multiple enlarged nodes around the hepatoduodenal ligament were noted, with no evidence of liver metastasis, ascites, or peritoneal metastases.

Following surgery, the patient was initiated on a daily dose of 60 mg prednisolone. Three days later, carbamazepine 200 mg and albendazole 400 mg were administered twice daily for 14 days. The steroid treatment continued throughout the antiparasitic therapy, followed by a rapid taper over a few days. The patient has now completed the albendazole therapy and is being monitored through multiple follow-up appointments. By his third visit, he had not developed any seizures. The histopathology report suggested findings consistent with distal cholangiocarcinoma (DCA).

Discussion

Del Brutto proposed a set of guidelines for the definitive diagnosis of cysticercosis. Patients can be diagnosed if they meet 1 absolute criterion, 2 major criteria, 1 major criterion, 2 minor criteria, and 1 epidemiologic criterion [3]. In our case, the

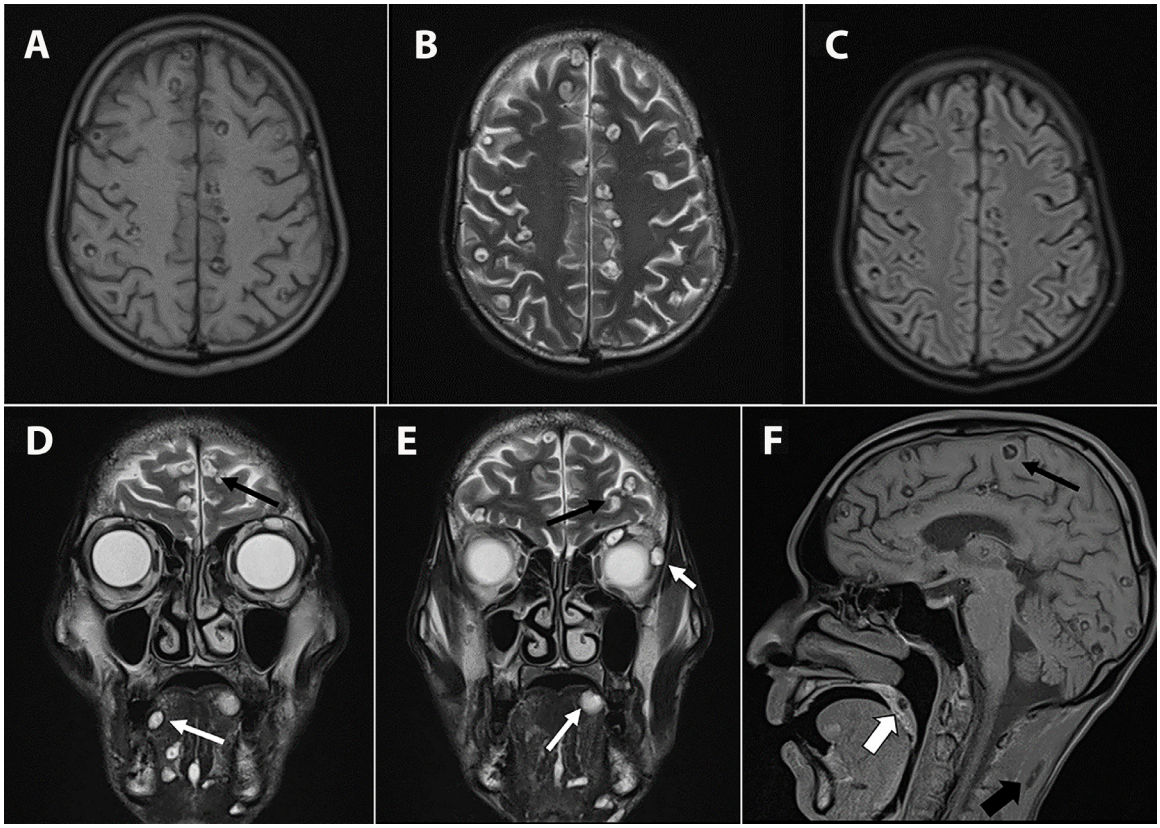

Fig. 7 – Magnetic resonance images of the brain: (A) T1 weighted axial image showing multiple iso intense lesions with central high signal intensity scolex in bilateral cerebral hemispheres. (B and C) T2 weighted and FLAIR axial images showing T2 high signal intensity cysts suppressed on FLAIR images with central scolex in bilateral cerebral hemispheres. (D and E) T2 weighted coronal images of the brain showing cysts in bilateral cerebral hemispheres (black arrows), tongue (long white arrows) and extra-ocular muscles (short white arrow). (F) T1 weighted sagittal image showing cysts in cerebral hemispheres (black arrows), soft palate (white boxed arrow) and trapezius muscle (black boxed arrow).

presence of cystic lesions with a visible scolex on CT or MRI confirmed the diagnosis of cysticercosis.

The engagement of multiple organs in DCC results in a diverse spectrum of manifestations. Central nervous system involvement can be either parenchymal (affecting the brain, spinal cord, or eye) or extra-parenchymal (involving the ventricles or subarachnoid space), or a combination thereof. This can lead to a range of symptoms including acute symptomatic seizures, headache, hydrocephalus, chronic meningitis, focal neurological deficits, psychological disorders, and dementia [4]. Imaging findings in our case revealed the involvement of the grey and white matter in the bilateral cerebral hemispheres, right cerebellar hemisphere, pons, and subarachnoid spaces. Despite these findings, none of the expected symptoms associated with such pathology were present.

Cardiac involvement in cysticercosis, particularly myocardial impairment, is uncommon and has been inadequately researched. It often remains asymptomatic, with diagnosis typically incidental during cardiac surgery or autopsy [5]. Our patient exhibited cardiac involvement indicated by granulomas in the myocardium and interventricular septum of the heart on CECT. Interestingly, both ECG and echocardiography were normal, highlighting the heightened sensitivity of CT in detecting cardiac lesions in cysticercosis.

Involvement of skeletal muscles leads to pseudohypertrophy of the affected muscles, typically without any accompanying tenderness [6]. In our case, the trapezius, paraspinal, and gluteal muscles were affected. Orbital cysticercosis may present with proptosis, inflammation, extraocular muscle involvement, subconjunctival cysts, lid nodules, and optic neuritis [6]. In this particular instance, only extraocular muscle involvement was observed without any additional symptoms.

According to a study by Delgado et al., oral cysticercosis most commonly affects the tongue, followed by the buccal mucosa, lower lip, and upper lip. Involvement of the tongue musculature by cysticercosis is common in swine but rare in humans, likely due to the high muscular activity and metabolic rate of human tongue muscles which may deter cysticercus lodgment and development in this location [7]. Moreover, in our case, the involvement of the soft palate was a notable finding.

The syndrome of DCC is characterized by pseudo muscular hypertrophy (100%), palpable subcutaneous nodules (87%), seizures (78%), and abnormal mentation (65%) [6]. Despite ex-

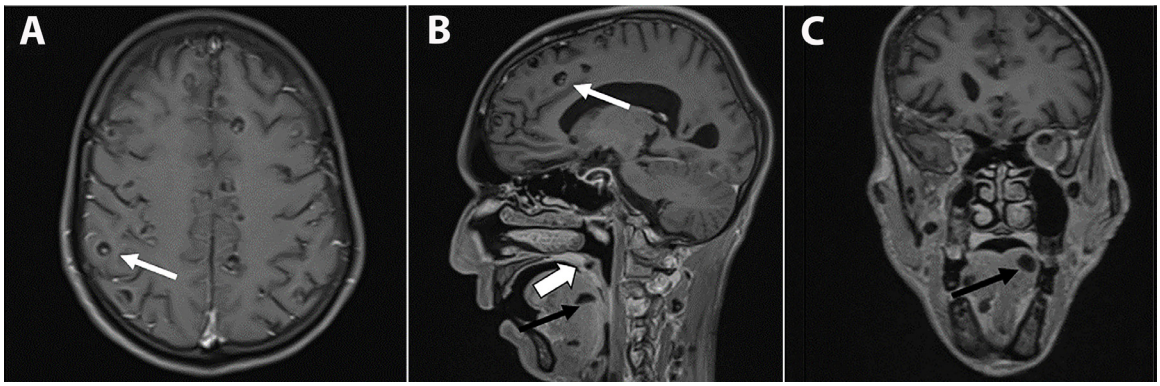

Fig. 8 – Post contrast axial (A), sagittal (B) and coronal (C) images showing mild peripherally rim enhancing lesions in bilateral cerebral hemispheres (white arrows), tongue (black arrows) and soft palate (white boxed arrow).

tensive dissemination, our patient had no symptoms except those caused by DCA.

Cysticercosis and neoplasia have been found together in previous studies, with prevalence rates ranging from 0.4% to 20.9% [8]. In our case, we found a DCA alongside disseminated cysticercosis. This aligns with a study by Cavellani et al., which found that tumors were mostly located in the gastrointestinal tract in patients with both neoplasia and cysticercosis. The literature is unclear about how cysticercosis and neoplasia occur together. One proposed mechanism suggests that cysticercus produces proteins that prevent the production of certain cytokines such as interleukin - 2,4 and interferon-gamma and the recruitment of macrophages, rendering the immune responses and proinflammatory cytokines ineffective in fighting tumors [8].

Conclusion

Disseminated cysticercosis can be completely asymptomatic despite its widespread dissemination. Although cysticercosis has been linked with other malignancies mentioned above, it has not been seen to be associated with periampullary cancers such as DCA. However, the concurrent presence of these 2 pathologies in this case suggests a potential association that warrants further investigation.

Patient consent

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent will be available for review if asked by the editor-in-chief of this journal.

Ethical approval

Patient information was de-identified and consent for publication has been obtained.

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