Introduction

A hydatid cyst is a parasitic disease that is very common in South Africa. The cestode, *Echinococcus granulosus* (*Taenia echinococcus*) is a parasite of the dog family (the definitive host) and the intermediate host is a variety of species of mammals such as sheep, cattle, goats, horses, pigs, and camels, while humans are accidental hosts who do not play a role in the biological cycle. Eggs from the cestode are usually ingested by man after contamination of the hands as a result of handling infested dog. Children are the usual victims, probably because they frequently place their hands in their mouths. The embryo reaches the liver through the portal venous system after penetrating the mucosa of the upper intestinal tract. About 70 per cent of hydatids lodge in the liver where they develop [1]. Those that pass through the liver are likely to travel via the right side of the heart to the lungs, which are second to the liver in frequency of involvement. However, in children, the primary site is the lung; mostly the right lung, especially the right lower lobe [2]. Finally, a few embryos pass through the lungs and via the systemic circulation, and lodge in other organs such as the brain, bones, or kidneys.

Occasionally, pulmonary hydatid cyst ruptures either into the pleural cavity or bronchus [3]. Rupture into the pleural cavity presents as pleural effusion and subsequent complications include secondary infection leading to empyema (in 7.6% cases), pleural thickening, collapsed lung, simple pneumothorax (in 2.4%–6.2% cases) [4] and tension pneumothorax [4], as well as dissemination and anaphylaxis [5]. In such patients, the diagnosis of hydatid cyst may at times be missed and the patient treated for other more common causes of such complications like pulmonary tuberculosis. In this communication, we present the rare case of a boy with ruptured hydatid cyst misdiagnosed as multidrug resistant tuberculosis (MDR TB) complicated by empyema at a peripheral clinic as this disease is endemic in South Africa.
Case Report

LS, a 10 year old boy was referred from a peripheral hospital in South Africa, to Nelson Mandela Academic Hospital (NMAH), Mthatha, with a possible diagnosis of MDR TB complicated by empyema. The patient had cough, malaise and general body weakness for six months. The cough was productive of yellowish sputum, sometimes blood stained with left chest pain aggravated by deep breathing and coughing. These were associated with loss of weight and loss of appetite; there was no history of night sweats. The mother was on treatment for pulmonary tuberculosis and all the six family members usually slept in one room.

A chest radiograph carried out by one of us (TIB) as showing features that were consistent with a left pleural effusion or collapse/consolidation of lower lobe left lung (Figure 1). Scanty pus recovered from a left pleuracentesis at the peripheral hospital was not analyzed by the laboratory because it was insufficient. Sputum smear was negative for acid fast bacilli, while GeneXpert TB test was negative for *Mycobacterium tuberculosis* complex. HIV Elisa test was non-reactive.

![Figure 1: Chest radiograph - PA view](image)

Figure 1: Chest radiograph - PA view (There is opacity in the left hemithorax sparing the left apical region. The heart and the lower part of trachea are displaced to the right side).

The patient had received anti-tuberculosis drugs (INH, rifampicin, pyrazinamide and ethionamide for 2 months, followed by INH and rifampicin for 2 months) in standard doses and defaulted for two months thereby failing to complete the six-month course. Hence a diagnosis of multidrug resistant pulmonary tuberculosis complicated by empyema was suspected.

On admission to the NMAH, he was severely wasted, weighed 23 kg (weight-for-age < -3 z-score), was afebrile (temperature, 37°C) with no significant peripheral lymphadenopathy. The respiratory rate was 28 breaths per minute with no signs of respiratory distress. His left hemithorax was prominent, the trachea was deviated to the right of midline, while percussion note was stony dull over the left hemithorax, with much reduced breath sounds over the left lower lobe. Cardiovascular findings were normal except for the apex beat which was visible and palpable just below the xiphisternum with heart sounds heard on the right side of the chest. Findings in other systems were within normal limits except a palpable liver of 2 cm below the right costal margin.

A repeat postero-anterior radiograph of the chest revealed similar findings as before, while a left lateral radiograph revealed no additional findings. A repeat pleural tap was dry. However, a chest ultrasound revealed a loculated pleural fluid collection with an echogenic capsule. The fluid was not thick and had no septum. Suggested radiological diagnosis was loculated left pleural fluid or a large cyst of uncertain aetiology. While further investigations were being carried out, he was restarted on daily oral rifampicin, 400 mg; INH, 400 mg; pyrazinamide, 800 mg; ethionamide, 400 mg; together with pyridoxine,12.5 mg; intravenous (i.v.) cloxacillin 500 mg q 6 h and i.v. gentamicin 100 mg daily.

Mantoux test was positive at 20 mm. *Echinococcus granulosa* ELISA revealed low positive titre of 1:64. Under i.v. midazolam (0.2 mg/kg) with xylocaine skin anaesthesia, a blunt dissection through the sixth left intercostal space at mid-axillary line was achieved and insertion of an ICD produced only about 50 ml pus. The drain was removed and re-inserted through the 8th intercostal space at mid-axillary line. This resulted in an additional 20 ml of pus being obtained, and this was followed by about 500 ml of blood stained purulent fluid, while the patient coughed out some more.

This was quickly followed by surgical emphysema at the site of tube insertion. The emphysema became worse, spreading all over the body except the upper part of the head and distal extremities. There was associated severe respiratory distress. A post ICD insertion chest radiograph (Figure 2) revealed a cavity in the left lower lobe with the ICD in situ. An atelectasis adjacent to the upper border of the cavity was identified, while a large mediastinal emphysema and shift of the heart to the right side were evident.

At this juncture, a possible bronchopleural fistula and ruptured tuberculous cavity was entertained. The SpO₂ was 99% (on 1 l/min of oxygen from a cylinder), and the arterial blood gases (ABG) revealed respiratory alkalosis (pH 7.53, PaCO₂ 22.9 mmHg, PaO₂ 123 mmHg, HCO₃⁻ 26.6 mmol/L, base excess 2 mmol/L).
Figure 2: Chest radiograph - PA view (There is a cavity in left lower lobe with atelectasis adjacent to the upper border of the cavity. A large mediastinal emphysema is noted. Consolidations in the right upper lobe, and severe chest wall and subcutaneous emphysema are evident).

The clinical condition worsened as evidenced by blood oxygen desaturating despite 5 l/min of oxygen, which was delivered via nasal prongs and clinical evidence of left pneumothorax was entertained. An urgent chest CT scanograph confirmed left pneumothorax, and a large cavity (containing hydatid endocyst) communicating with the left bronchus and the subcutaneous tissue, as well as extensive chest wall, subcutaneous tissue, mediastinal and spinal canal emphysema (Figure 3).

Figure 3: CT of the chest - coronal view (There is a large left lung pericyst with collapsed endocyst. Communication between the pericyst and chest wall through the ICD path. Large chest wall, subcutaneous, mediastinal and spinal canal emphysema are present).

The initial antibiotics were replaced with metronidazole (250 mg 8 hourly), cefotaxime (500 mg 6 hourly) and amikacin (400 mg daily) given intravenously as well as oral praziquantel 300 mg 8 hourly for hydatidosis. About 48 hours after the diagnosis, findings at left thoracotomy included ruptured dead hydatid cyst involving the posterior left lower lobe that was compressed to form an ectocyst. Surrounding the ectocyst was a margin of atelectatic lung tissue. Also communicating with the cavity of the ectocyst were four bronchial and bronchiolar openings or fistulae. Surgical procedure included cysts removal, individual closure of each fistula with 3/vicryl and then capitation of the cavity. Thorough washing of the thoracic cavity with normal saline was carried out while two chest drains, a basal and an apical, were left in situ. Expectedly, sections of the endocyst showed characteristic fragments of laminated, amphophilic wall of the hydatid cyst.

The patient was transfused postoperatively with 150 ml of concentrated red cells. Chest radiograph on the 5th postoperative day showed that the right lung was normal. There was a residual cavity in the left suprarenal region posterior to the heart, otherwise the left lung was normal. A small pleural effusion was evident but the mediastinum had returned to the midline.

He was discharged home, on 15th postoperative day, on praziquantel 300 mg 8 hourly for one month. At a follow-up clinic three weeks after discharge, he had gained 2 kg in weight, there were no complaints and chest findings were within normal limits. The patient denied ever coming in close contact with dogs, but accepted playing with a sheep in a distant neighbour’s house.

Discussion

Hydatid disease (echinococcosis) does not present a constant clinical pattern, and as it is so often the case, lack of high index of suspicion rather than lack of knowledge about it accounts for most of the misdiagnoses. Our patient denied ever coming in contact with dogs, but indicated playing with a sheep in the home of a neighbour. Thus, the probable source of infestation in this case was a sheep harbouring the eggs on the fur, which might have occurred while grazing on contaminated ground even though this has not been previously described. It is also more likely that our patient failed to recall an encounter with a dog.

The clinical diagnosis of pulmonary tuberculosis in our patient was relevant because there was a history of chronic cough associated with weight loss, haemoptysis and contact with a tuberculosis case. A positive Mantoux test confirmed that the patient was infected with the M. tuberculosis, but this is not a proof of disease. However, the lack of laboratory confirmation of tuberculosis with the sensitive GeneXpert Tb test did not persuade us to look for an alternate diagnosis because childhood tuberculosis is paucibacillar.
Haemoptysis in pulmonary hydatid disease is a common presenting symptom. Mechanisms include pressure erosion of a bronchus, obstructive infection, and cyst rupture or very rarely, erosion of a major vascular structure. However, symptomatic hydatid disease of the lung, more often follows rupture of the cyst [6,7], which may occur spontaneously or as a result of trauma or secondary infection. Repeated diagnostic pleuralcentesis and secondary infection were the most likely cause of the rupture in our patient. In a contained rupture, only the endocyst is torn and the contents of the cyst are contained by the pericyst. In a communicating rupture, the contents of the cyst escape into the tracheobronchial tree through bronchioles that have been incorporated into the pericyst. Direct rupture into the pleura follows tearing of both the endocyst and the pericyst, with discharge of the contents of the cyst directly into the pleural cavity [8]. Our patient had direct rupture during presentation accounting for clinical features of loculated empyema; and subsequently the ICD led to the bronchopleural fistula, pneumothorax and extensive surgical emphysema.

Albendazole® is usually given pre- and post-operatively in the management of ruptured pulmonary hydatid cyst. However, this drug was not available in our hospital. Praziquantel was given instead, even though it has not been so well studied but shows promise. Promotion of hand hygiene among children should reduce infestation with the eggs of the cestode and prevent hydatidosis.

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References