The Great Mimicker: A case of ruptured lung cyst

Abstract

Introduction: One of the most prevalent species, Echinococcus granulosus, is the parasite zoonotic infection that causes echinococcosis, also known as hydatid disease (HD). This species usually affects the liver, lungs, and, in rare cases, the kidney, muscles, bones, and spleen. Humans can only contract it through close contact with animals. Due to overlapping characteristics, pulmonary hydatid cysts might be mistakenly diagnosed as pneumonia in endemic areas like India, where tuberculosis (TB) is prevalent. A definitive diagnosis of HD can be established with the aid of radiological and pathological results.

Case Presentation: A 60-year-old male from Munsyari, Uttrakhand, presented with a 3-month history of breathlessness, cough, and expectoration. Despite being on anti-tubercular therapy (ATT) for two months which was advised by local practioner based on his clinical features, the chest X-ray opacities showed no improvement. Computed tomography (CT) revealed a 5.3 x 4.5 x 6.1 cm hypodense lesion in the right upper lobe with floating membranes and an "air bubble sign," suggesting a ruptured hydatid cyst. A second cyst was identified in the liver. However patient had no travel history in recent days but a thorough history revealed that patient was a pet dog owner which is a well known risk factor for *E. granulosus* and thus causing echinococcosis.

The patient was treated with Albendazole and underwent a cystostomy with capitonnage via thoracotomy. The sample was later sent for gold standard histopathology and a final diagnosis of hydatid cyst was given.

Conclusion: This case highlights the importance of considering hydatid disease in differential diagnoses for non-resolving pneumonia or suspected TB, particularly in high-altitude, endemic regions.

Keywords: Anti-tubercular, Tuberculosis, pulmonary hydatid cyst, pneumonia

Introduction

Hydatid disease is a parasitic infestation caused by Echinococcus granulosus characterized by cystic lesions in the liver and lungs but rarely in other parts of the body [1,2]. E. granulosus causes cystic echinococcosis, which has a worldwide distribution. Humans are exposed less frequently to other species like E. multilocularis, which causes alveolar echinococcosis. E. vogeli and E. oligarthrus are rare species and cause polycystic echinococcosis. [3] In cystic echinococcosis (CE), humans are an accidental host and are usually infected by handling an infected dog. The liver and lungs are the most frequently involved organs. Pulmonary disease appears to be more common in younger individuals. Although most patients are asymptomatic, some may occasionally expectorate the contents of the cyst or develop symptoms related to compression of the surrounding structures.[3] CE is endemic in many parts of the world, particularly the Mediterranean countries, Central Asia including the Tibetan Plateau, Northern and Eastern Africa, Australia, and South America. [4]

Case presentation

A 60-year-old man from Munsyari, Uttarakhand, presented to us with complained of dyspnea, coughing with expectoration for three months. His Chest X-ray (CXR) PA view (Figure 1) showed large homogenous opacity in the right middle zone and inhomogeneous opacities in the right upper zone. Based on these clinical symptoms and radiological investigations, the patient was clinically diagnosed elsewhere with pneumonia caused by tuberculosis and he was started on anti-tubercular therapy (ATT) for same for two months. However, when a second CXR was done in our hospital it revealed no improvement when compared to the first CXR. Because of this clinico-radiological picture, sputum for acid fast bacilli (AFB) and cartridge based nucleic acid amplification test (CBNAAT) were done which came out to be negative. So a CT chest(Figure 2)was performed which showed a well defined rounded area of hypo density measuring 5.3x 4.5x6.1 cm(APxTRxCC) involving anterior segment of right upper lobe. Internally, air specks were noted showing communication with segmental bronchus. On post contrast study curvilinear enhancing floating membranes were noted within the lesion. This picture lead to the possibility of hydatid lung disease with rupture into the bronchus and surrounding inflammation in right lung parenchyma. Another hydatid cyst was noted in segment vii of right lobe of liver. History was again reviewed and patient confirmed domestic animals especially a pet dog. Serology for hydatid cyst was also done though it came to be negative. Therefore, a final diagnosis of hydatid cyst of lung with communicating rupture into bronchus along with hydatid cyst in liver was made.

Following diagnosis patient was discharged with twice daily tablet Albendazole for 3 months. And later on, the patient underwent thoracic surgery for cystostomy with capitonnage at different centre (Figure 3). Intercostal drainage (ICD) tube was inserted after the procedure which was later removed (Figure 4,5). The excised specimen was sent for gold standard histopathology which finally confirmed the clinico-radiological diagnosis of hydatid cyst.

Discussion

Hydatid disease is a parasitic infestation caused by Echinococcus granulosus. The lungs are the second most common sites for hydatid cysts after the liver [1,2]. The majority of lung hydatid cysts are silent and either small or medium in size. Non-complicated hydatid cysts are usually discovered incidentally during routine chest X-rays for complaints other than chest diseases [5]. Giant hydatid cysts and complicated cysts, on the other hand, are usually symptomatic. The common presentations are compression symptoms such as a dry cough in cases of very large cysts; a productive cough in cases associated with communication with the bronchial tree; and chest pain and dyspnoea in the case of rupture to the pleural cavity [6]. Anaphylactic shock is a rare presentation (seen in cases of rupture to the pleural cavity). The diagnosis is easy in endemic areas. The patient is usually in good general health in cases of non-complicated cysts and chest X-ray will show a well-circumscribed dense homogenous opacity. A water-lily radiological sign is a diagnostic feature for a cyst associated with communication with small bronchioles and with a detached laminated membrane. Productive cough of grape skin-like material is diagnostic in ruptured hydatid cysts communicated with medium sized bronchioles [7].

Pulmonary hydatid cysts is treated by pharmacotherapy and/or surgery. Surgical intervention is the treatment of choice though pharmacotherapy may also be useful in selected patients. Medical therapy of pulmonary hydatid cyst includes benzimidazoles group of drugs, for example, mebendazole (MBZ) or albendazole (ABZ).

ABZ also achieves a higher plasma and intracystic drug concentration. Its plasma concentration is 10–40 times higher than that of MBZ [8]. ABZ requires a minimum contact period of 11 days to have a significant response [9]. The usual recommended a dosage of ABZ is 10–15 mg/kg/day, taken twice daily, and that of MBZ is 40–50 mg/kg/day, thrice daily. Fat rich meals increase the bioavailability of the drugs [10]. The optimal duration of pharmacotherapy in pulmonary hydatidosis is not known, but it should be given for a minimum period of 3–6 months.

For manangement surgery is indicated in following cases, large cysts that are superficial and likely to rupture, infected cysts, cysts in vital anatomical locations, and cysts exerting substantial mass effect [11]. The goal of surgical intervention includes removal of the entire cyst while preserving the lung parenchyma as much as possible and without allowing intraoperative spillage. Various surgical techniques available include enucleation, pericystectomy, cystostomy with capitonnage, open aspiration, and lung resection [12]. Prevention of intraoperative spillage can be achieved by placing gauze soaked with a hypertonic saline solution (20%) or povidone-iodine solution [13].

Conclusion and clinical significance

In regions where tuberculosis is endemic, pulmonary hydatid disease is rare and frequently misdiagnosed, leading to the inappropriate initiation of anti-tubercular therapy (ATT). This case

highlights the need for clinicians to maintain a high index of suspicion for *Echinococcus granulosus* when patients from high-altitude areas do not respond to ATT. To avoid diagnostic delays, a thorough review of domestic animal exposure and a careful search for specific radiological signs, such as the "air bubble" or "water-lily" signs, are essential before committing to long-term tuberculosis treatment. ATT in these cases should only be started by a pulmonologist after ruling out other diagnostic possibilities and frequent follow up.

References

1. Kavukcu S, Kilic D, Tokat AO, Kutlay H, Cangir AK, Enon S, Okten I, Ozdemir N, Gungor A, Akal M, Akay H: Parenchyma-preserving surgery in the management of pulmonary hydatid cysts. J Invest Surg 2006, 19(1):61-68.

2. Safioleas M, Misiakos EP, Dosios T, Manti C, Lambrou P, Skalkeas G: Surgical treatment for lung hydatid disease. World J Surg 1999, 23(11):1181-1185

3. Mandal S, Mandal MD. Human cystic echinococcosis: Epidemiologic, zoonotic, clinical, diagnostic and therapeutic aspects. Asian Pac J Trop Med 2012;5:253-60..

4. Wahlers K, Menezes CN, Wong ML, Zeyhle E, Ahmed ME, Ocaido M, et al. Cystic echinococcosis in sub-Saharan Africa. Lancet Infect Dis 2012;12:871-80.

5. Robert ES, Eugene JM, William FM, Sally HE, Stacey M: Case records of the Massachusetts General Hospital. Weekly clinicopathological exercises. Case 29-1999. A 34-year-old woman with one cystic lesion in each lung. N Engl J Med 1999, 341(13):974-982

6. Saidi F: Treatment of Echinococcal cysts. In Mastery of Surgery 3rd edition. Edited by: Nyhus LM, Baker RJ, Fisher JE. Boston, New York, Toronto, London: Little, Brown & Co; 1997:1035-1052.

7. Beggs I: The radiology of hydatid disease. AJR Am J Roentgenol 1985, 145(3):639-648.

8. Morris DL, Gould SE. Serum and cyst concentrations of mebendazole and flubendazole in hydatid disease. Br Med J (Clin Res Ed) 1982;285:175

9. Taylor DH, Morris DL, Richards KS. Echinococcus granulosus: In vitro maintenance of whole cysts and the assessment of the effects of albendazole sulphoxide and praziquantel on the germinal layer. Trans R Soc Trop Med Hyg 1989;83:535-8.

10. Guidelines for treatment of cystic and alveolar echinococcosis in humans. WHO Informal Working Group on Echinococcosis. Bull World Health Organ 1996;74:231-42.

11. McManus DP, Zhang W, Li J, Bartley PB. Echinococcosis. Lancet 2003;362:1295-304.

12. Nabi MS, Waseem T. Pulmonary hydatid disease: What is the optimal surgical strategy? Int J Surg 2010;8:612-6.

13. Koul PA, Singh AA, Ahanger AG, Wahid A, Sofi BA. Optimal duration of preoperative anti-helminthic therapy for pulmonary hydatid surgery. ANZ J Surg 2010;80:354-7.

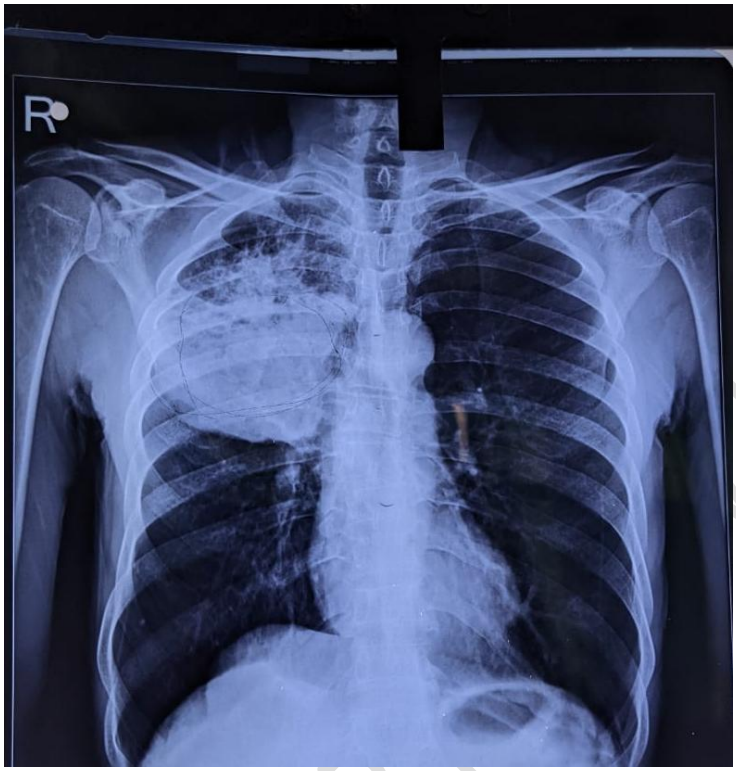

Figure 1. CXR PA view shows large homogenous opacity in the right middle zone and inhomogeneous opacities in the right upper zone

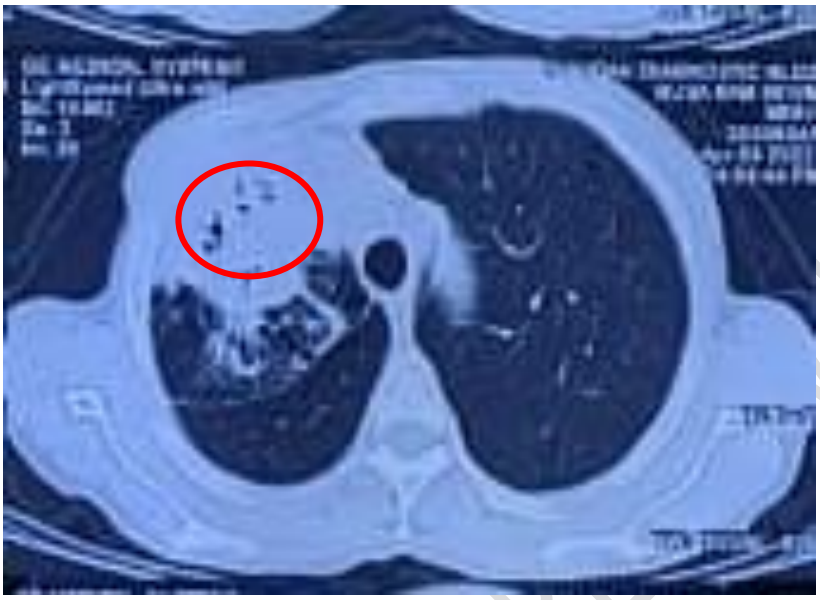

Figure 2. CT chest (lung window) small air bubbles within the perforated pulmonary cysts named as “air bubble sign”

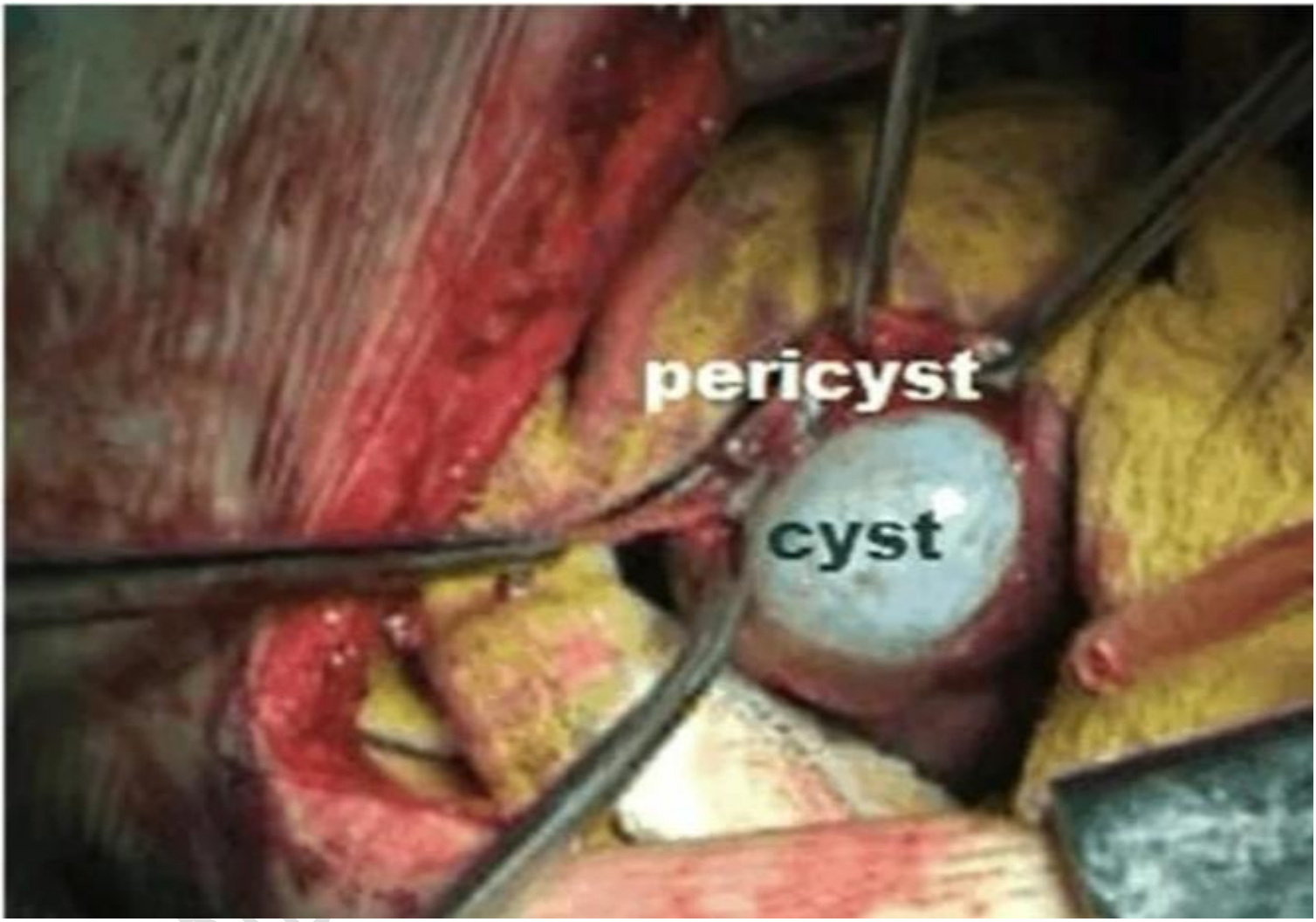

Figure 3. Post operative excised macroscopic appearance of Hydatid cyst of lung

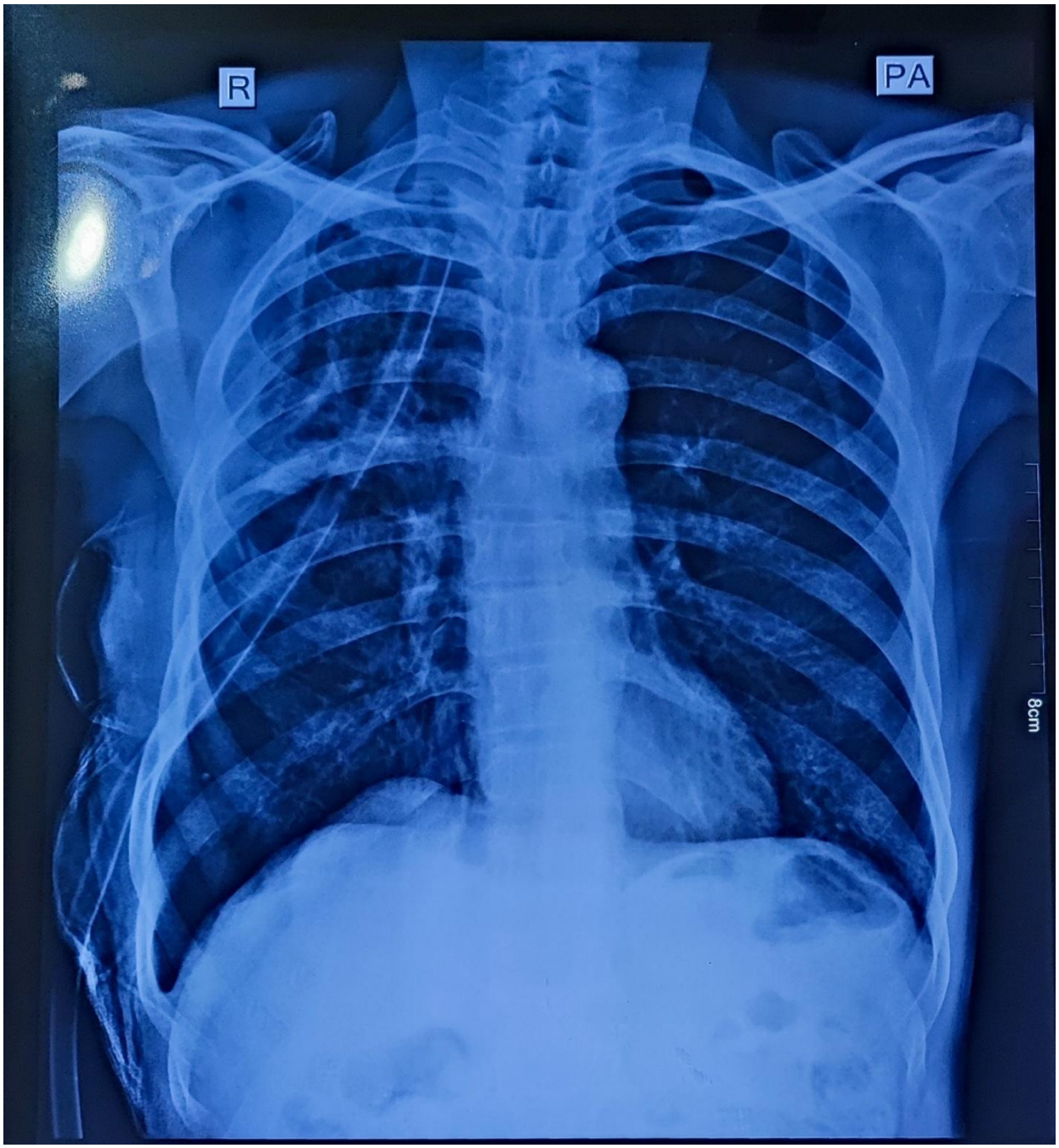
R
PA
8cm

Figure 4. CXR PA view of the same patient after removal of hydatid cyst. An ICD on the right side was inserted after the procedure.

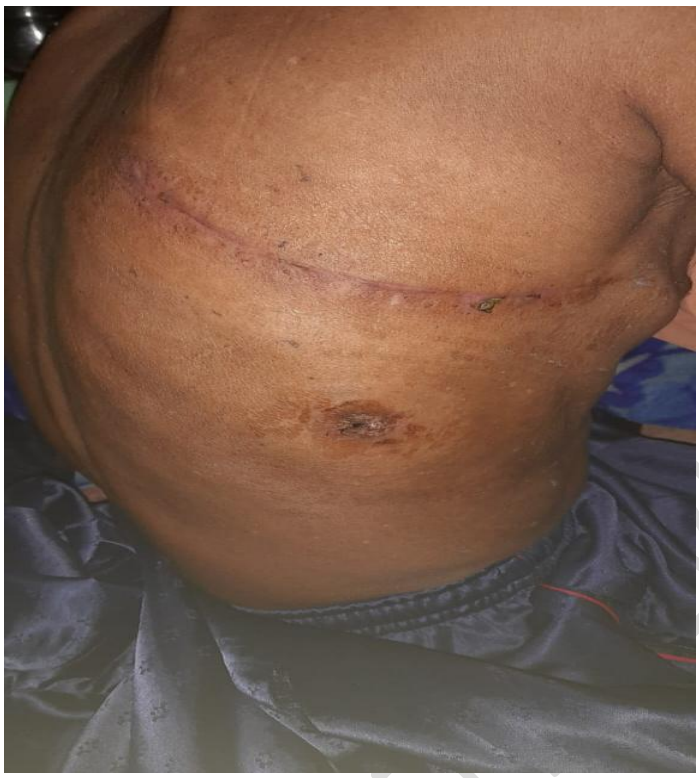

Figure 5. Thoracotomy mark (curvilinear mark along the rib) and ICD tube mark (small round mark below)

199

200