

## REVIEW ARTICLE

# Spontaneous Biliary Pericardial Tamponade: A Case Report and Literature Review

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**Abstract: Background:** Biliary pericardial tamponade (BPT) is a rare form of pericardial tamponade, characterized by yellowish-greenish pericardial fluid upon pericardiocentesis. Historically, BPT reported to occur in the setting of an associated pericardiobiliary fistula. However, BPT in the absence of a detectable fistula is extremely rare.

**Learning objective:** A biliary pericardial tamponade is a rare form of tamponade warranting a prompt workup (e.g., MRCP or HIDA scan) for a potential fistula between the biliary system and the pericardial space. A pericardio-biliary fistula can be iatrogenic or traumatic. People with a history of chest wall trauma, abdominal surgery, or chest surgery are at increased risk. The use of HIDA scanning plays a salient role in effectively surveilling for the presence of a fistula – especially when MRCP is contraindicated.

**Case Presentation:** A 75-year-old Hispanic male presenting with dyspnea and diagnosed with cardiac tamponade is the subject of the study. Subsequent pericardiocentesis revealed biliary pericardial fluid (bilirubin of 7.6 mg/dl). The patient underwent extensive workup to identify a potential fistula between the hepatobiliary system and the pericardial space, which was non-revealing. The mechanism of bile entry into the pericardial space remains to be unidentified.

**Literature Review:** A total of six previously published BPT were identified: all were males, with a mean age of 53.3 years (range: 31-73). Mortality was reported in two out of the six cases. The underlying etiology for pericardial tamponade varied across the cases: incidental pericardio-biliary fistula, traumatic pericardial injury, and presence of associated malignancy. -

**Conclusion:** Biliary pericardial tamponade is a rare form of tamponade that warrants a prompt workup (e.g., Hepatobiliary Iminodiacetic Acid – HIDA scan) for an iatrogenic vs. traumatic pericardio-biliary fistula. As a first case in the literature, our case exhibits a biliary tamponade in the absence of an identifiable fistula.

**Keywords:** Pericardial effusion, cardiac tamponade, biliary cardiac effusion, pericardiobiliary fistula, BPT, pericardiocentesis.

## 1. INTRODUCTION

The occurrence of biliary tamponade is exceedingly rare and has only been reported a few times in the literature. The reported cases involved traumatic or surgical manipulation of organ systems within proximity to the heart. Biliary tamponade was also reported in association with metastatic malignancies. All reported biliary tamponade cases were

in male patients; however, no prior hypothesis was raised to explain this potential sex-specific predilection. The presentation and management of biliary tamponade are identical to other types of cardiac tamponade. Mortality was verified in a couple of the reported cases [1, 2]. The aim of this case report with literature review is to highlight this rare diagnosis and the demographics, potential risk factors, and workup for this rare type of cardiac tamponade.

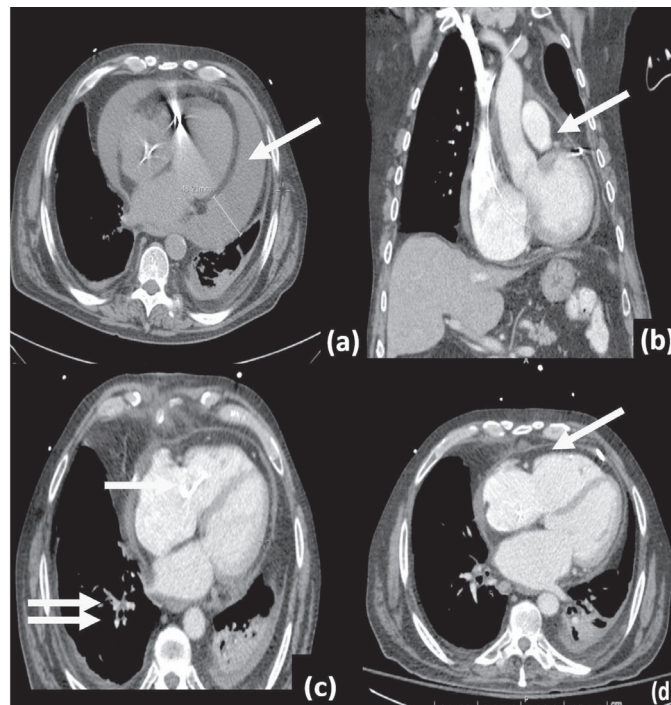
## 2. CASE REPORT

This is a 75 - year - old Hispanic male patient with a history of paroxysmal atrial fibrillation on anticoagulation, im-

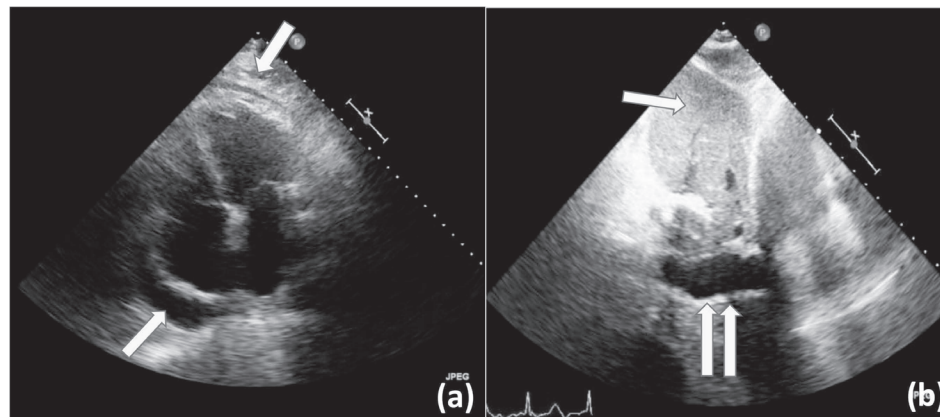
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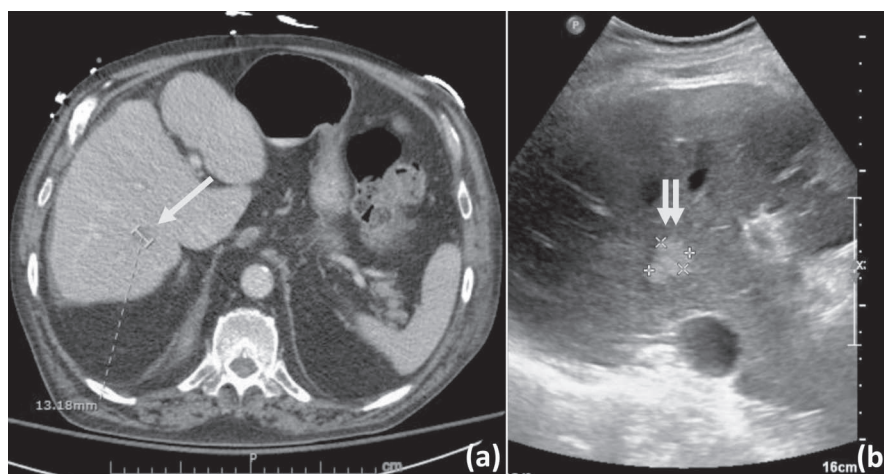
**Fig. (1).** (a) CT scan of the chest (prior to pericardiocentesis) showing moderate to large pericardial effusion (arrowed). (b) Contrast-enhanced chest CT scan (day 1 following pericardiocentesis) revealed persistent pericardial effusion (arrowed) as well as pericardial enhancement suggestive for the inflammatory or infectious process; no malignant lesion identified. (c) Calcified subcarinal lymph node (single arrow) and hypodense lesion in the liver (doubled arrows) could also be identified. (d) Repeat chest CT scan at one-month post-pericardiocentesis showing minimal reaccumulating pericardial fluids and trace pleural effusions. The pericardial air has resolved. (A higher resolution / colour version of this figure is available in the electronic copy of the article).



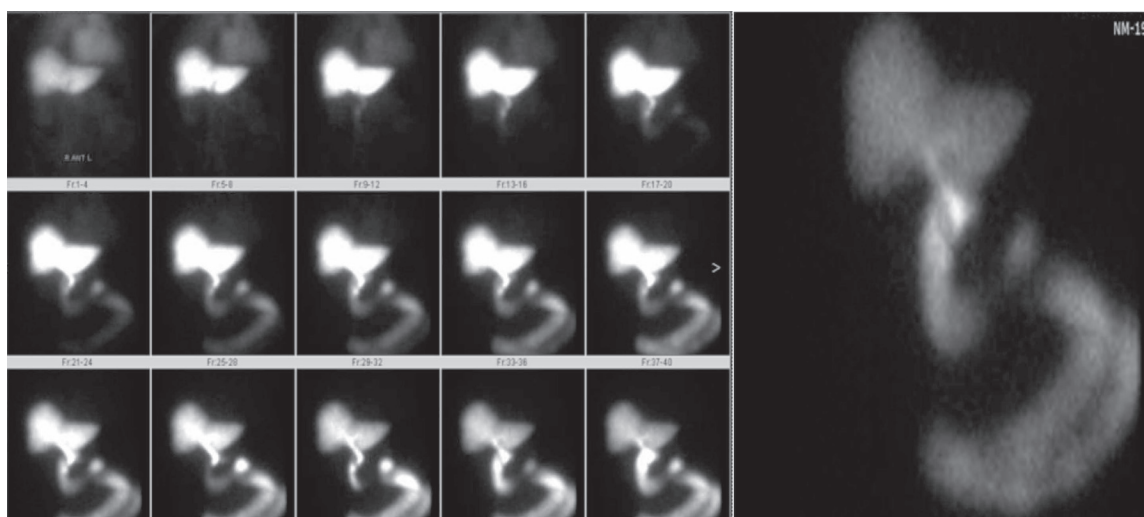
**Fig. (2).** (a) Transthoracic Echocardiography (TTE) showing the apical 4-chamber view with large circumferential pericardial effusion (yellow arrow) and echodense layer on along the visceral surface of the pericardium representing pericardial fat, soft tissue or fibrin deposits. (b) Subcostal view showing the inferior vena cava (double arrows) to be dilated with less than 50% inspiratory collapse at the level of hepatic veins (single arrow points to the liver). (A higher resolution / colour version of this figure is available in the electronic copy of the article).

planted cardiac pacemaker, colorectal carcinoma (with partial colectomy), and vestibular schwannoma. The patient presented to the emergency complaining of acute substernal chest pain and shortness of breath. The patient described his chest pain to be central that increases with lying down, and decreases with leaning forward. The patient denied any recent traumatic event. Physical examination revealed diminished precordial heart sounds and markedly elevated Jugular Venous Pressure (JVP). Vital signs revealed a blood pressure of 95/70 mmHg, heart rate of 82 BPM, respiratory rate of 19/minute, and temperature of 98.2°F (36.8°C).

Subsequently, a chest x-ray was obtained, which showed enlarged cardiac silhouette with bilateral pleural effusions. CT scan of the chest revealed moderate-to-large pericardial effusion (Fig. 1, 1a). Transthoracic Echocardiography (TTE) was performed next, which revealed an echo dense layer of a large circumferential pericardial effusion (Fig. 2a). The inferior vena cava was also shown to be dilated with less than 50% inspiratory collapse (Fig. 2b). These physical examination and radiological findings were diagnostic for cardiac tamponade.



**Fig. (3).** (a) Dedicated abdominal CT scan showing a small (1.3 cm in diameter) low-density lesion in the liver (arrowed) of uncertain etiology. (b) Right upper quadrant ultrasound showing a hypodense echogenic liver lesion measuring 1.8 cm (arrowed). (A higher resolution / colour version of this figure is available in the electronic copy of the article).



**Fig. (4).** Hepatobiliary Iminodiacetic Acid (HIDA) scan performed with an infusion of technetium 99m labeled Choletec showing a rapid clearance of blood pool activity indicating normal hepatic function. At 1 hour, the liver is essentially cleared. While bile duct and small bowel activities are present, there is no evidence of activity within the pericardium. There is no activity seen within the gallbladder, which is surgically absent (*i.e.*, cholecystectomy). There is no evidence of a biliary system to the pericardial fistula. (A higher resolution / colour version of this figure is available in the electronic copy of the article).

Next, interventional radiology service was consulted and a right-sided subxiphoid pericardiocentesis was performed after one trial with pericardial drain insertion. The procedure lasted for about 20 minutes and a total of one liter of non-hemorrhagic greenish-yellowish bile-like fluid was drained. Analysis of pericardial fluid revealed an exudative effusion, bilirubin level of 7.6 mg/dl, amylase level of 23 U/L, lipase level of 14 U/L, WBC count of 29,000 cells/ $\mu$ L, RBC count of 3,000 cells/ $\mu$ L, glucose level < 2 mg/dl, LDH level of 1,463 U/L, and Adenosine Deaminase of 2.6 mg/dl. The pericardial fluid culture was negative for bacterial and fungal infections. Pericardial fluid cytology was also negative for malignant cells. Laboratory tests showed normal liver functions, kidney functions, and thyroid panel. A nasopharyngeal viral panel was also negative. Additionally, Interferon Gamma Release Assay (IGRA) and 16S ribosomal study were negative. The Erythrocyte sedimentation rate was elevated at 64 mm/hour.

A follow-up CT of the chest following pericardiocentesis revealed partial resolution of pericardial effusion and pericardial enhancement, which might reflect an underlying inflammatory or infectious process. The imaging did not identify any malignant lesions; however, it showed calcified subcarinal lymph nodes and hypodense lesion in the liver (Fig. 1b and 1c). A dedicated abdominal CT scan showed a small (1.3 cm in diameter) low-density lesion in the liver of uncertain etiology (Fig. 3a) and right upper quadrant ultrasound showed an ovoid echogenic lesion measuring 1.8 cm (Fig. 3b).

Further workup to identify the source for the biliary pericardial effusion included: (1) Right Upper Quadrant (RUQ) ultrasound, and (2) Hepatobiliary Iminodiacetic Acid (HIDA) scanning. RUQ ultrasound showed a 1.8 cm echogenic liver lesion consistent with cavernous hemangioma but was otherwise normal. Further, HIDA scanning did not re-

**Table 1. Cases of biliary cardiac tamponade reported in the literature: summary and outcomes.**

-	Age/Gender	Diagnostic Studies	Diagnostic Studies Result	Outcome
Marlatt, 1991 [1]	48/Male	Barium swallow, CT chest, HIDA scan, upper GI series, V/Q scan	Incidental pericardial fistula	Dead
Kotoulas, 2003 [2]	66/Male	Chest X-ray, CT of the chest, TTE	Metastatic lung adenocarcinoma, hepatic hydatid cyst	Dead
Chavez, 2018 [3]	31/Male	Chest CT angiography, abdomen/pelvis CT, HIDA	Pericardiobiliary fistula	Survived
Lee, 2013 [4]	68/Male	TTE, Chest CT scan	Biliary tamponade secondary to biliary stent migration; pericardiocentesis & stent retrieval <i>via</i> ERCP	Survived
Surman, 2013 [5]	73/Male	Abdominal CT, chest CT, echo, fluoroscopy	Percutaneous pericardial injury	Survived
Von Riedenauer, 2010 [6]	34/Male	CT, HIDA, ERCP	Fibropurulent pericarditis	Survived

veal any fistula connecting the biliary system to the pericardial space (Fig. 4). The patient had an implanted cardiac pacemaker that was not compatible with Magnetic Resonance Cholangiopancreatography (MRCP), and hence, HIDA scanning was the imaging modality of choice to assess for a fistula involving the biliary system.

Due to the high output-flow from the pericardial drain, a decision was made to perform a pericardial window and drain procedure. During the pericardial window, a biopsy specimen was also obtained from the pericardium, which showed chronic pericarditis with fibrosis of the pericardial wall – no evidence of malignancy. The smear obtained during the procedure was also negative for acid-fast bacilli staining. In the meanwhile, the pericardial drain continued to be actively draining from the pericardial space. Subsequently, a repeat chest CT scan was performed after the procedure showed a left-approach pericardial drain extending into the right aspect of the pericardial space with a small residual pericardial fluid – no frank blood was noticed. Follow-up TEE 2 days after the procedure showed near resolution of pericardial effusion. Thereafter, the patient commenced significant clinical improvement with complete resolution of his chest pain and normalization of vital signs. The patient continued to have shortness of breath that is likely attributed to collapse in the left lower lung as a complication for his course of hospitalization. The patient was eventually discharged with nearly complete recovery after 3 weeks of hospitalization. The pericardial drain continued to be in place throughout the admission time until it was removed one day prior to discharge at week three of hospitalization.

Close follow-up with cardiology in the outpatient settings was scheduled. The patient continued to be symptoms-free at the one-month follow-up visit. A repeat chest CT (Fig. 1d) at one month showed minimal amount of pericardial fluid, resolution of pericardial air, and trace pleural effusions.

### 3. DISCUSSION

Biliary pericardial effusion is a rare form of pericardial effusion that occurs in patients with predisposing conditions, such as surgical manipulation of the chest/abdominal content or penetrating trauma that establishes a fistula between the

biliary tree and the pericardium [3-5]. The report of biliary effusion in the pericardial space in the absence of these known risk factors is even exceedingly rare.

We hereby report on a case of biliary tamponade with no identifiable etiology. However, of all comorbidities, the patient's history of colon cancer with possible micro-metastasis sounds to be the most relevant. The possibility of malignancy causing micro-metastases to the pericardial space cannot be ruled out, even with negative pericardial fluid cytology and pericardial wall biopsy. While the patient's workup included chest, abdominal, and pelvic CT scans. No further investigative workup (*e.g.*, PET-CT scan or gallium scintigraphy) for primary or metastatic malignancy was performed. A second hypothesis is that 'micro-fistulas' under the threshold of detection by HIDA scan may have occurred – allowing for leakage of bile into the pericardial space. Even though our patient underwent HIDA scanning twice one week apart, no fistula could be identified. Lastly, a less appealing hypothesis is the presence of chronic micro-bleeding into the pericardial space giving rise to bilirubin via chronic hemolysis. The RBC count upon pericardiocentesis was 3,000 cells/ul; however, this can also be attributed to the traumatic nature of the procedure. Finally, the possibility of bacterial or fungal infection has theoretically been ruled in lieu of the negative fluid culture and negative pericardial biopsy. Nonetheless, the possibility of a viral infection not detected by the viral panel cannot be excluded.

In December 2019, a systematic search was conducted using PubMed and Google Scholar to identify biliary effusion cases reported in the literature. Data reviewed included demographics, surgical and trauma history, imaging studies (*e.g.*, TTE, HIDA scanning), and treatment outcomes. A total of six biliary pericardial tamponade cases were identified. The pertinent findings are summarized in Table 1 [1–6]. The mean age was 53.3 years (range: 31–73 years). All the six patients were males. Four cases received emergent pericardiocentesis as part of biliary tamponade management. Death was the outcome in two of the six cases [1, 2]. The first biliary tamponade case reported in the literature goes back to 1991 [1]. The study demonstrated a pericardio-biliary fistula formation incidentally during HIDA scan evaluation for a patient with biliary vomiting after resection of esophageal

adenocarcinoma [1]. The presence of a fistula in such a case clearly explains the influx of bile into the pericardial space.

Furthermore, all the other five reported cases had an explanatory mechanism for the development of biliary tamponade. Kotoulas *et al.*, reported a case of biliary effusion as the initial manifestation of malignancy that coexisted with echinococcal hydatid cyst disease [2]. Hydatid cyst was suspected to be the driving force behind fistulization [2]. The case by Chavez *et al.*, attributed a tamponade to penetrate trauma from a gunshot wound [3]. Another case presented with biliary tamponade because of stent migration into the pericardial space [4]. During CT-guided needle biopsy of a liver mass, puncture of the pericardium precipitated the formation of the fistula. Lastly, additional two cases demonstrated iatrogenic intervention as the cause of biliary tamponade [5, 6]. All previously reported cases had adequate explanations for bile accumulation in the pericardial space; our case does not have a definitive explanation even though the pericardiocentesis and the subxiphoid puncture might have injured the liver creating acute connection from a biliary duct to pericardial space. Hence, the biliary tamponade in our case can be described as “spontaneous”.

Management of biliary tamponade is identical to that of non-biliary tamponade. Early pericardiocentesis should be undertaken when the classic triad for tamponade exists, as urgent aspiration of pericardial fluid to achieve hemodynamic stability is more crucial than a confirmatory diagnosis. In our review, four out of six cases received urgent pericardiocentesis. Mortality occurred in one-third of the cases [1, 2]. In addition to early pericardiocentesis, in biliary effusion, workup for underlying etiologies for biliary influx, with surgical repair in cases of fistulization, should also be undertaken. In our case, we could not identify a fistula that needs to be repaired.

## CONCLUSION

Biliary tamponade is a rare form of tamponade that is classically linked to the presence of pericardiobiliary fistula. The reported mechanisms for fistulization include iatrogenic and traumatic injuries. Biliary tamponade was also reported in association with malignancies. The presence of biliary effusion warrants a prompt workup for pericardiobiliary fistula (MRCP or HIDA scanning). Even though we called our

case spontaneous biliary pericardial tamponade because all imaging and investigations did not reveal a clear underlying cause but malignancy, pacemaker or pericardiocentesis procedure can be hypothesized as possible underlying etiologies.

## CONSENT FOR PUBLICATION

Not applicable.

## FUNDING

None.

## CONFLICT OF INTEREST

The authors declare no conflict of interest, financial or otherwise.

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Declared none.

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