IDCases 37 (2024) e02036

Contents lists available at ScienceDirect

IDCases

journal homepage: www.elsevier.com/locate/idcases

Case report

Ultrasound findings of Fitz-Hugh-Curtis Syndrome (FHCS) associated with splenic tuberculosis in an HIV-positive male patient

Ibrahima Niang [a,b,*,1], Daouda Thioub [c], Mamadou Ly [a], Abdourahmane Ndong [d], Fallou Galass Niang [e], Abdoulaye Dione Diop [a,b], Sokhna Ba [a,b]

[a] Radiology department, Fann University Hospital Center, Dakar, Senegal
[b] Radiology department, Cheikh Anta Diop University, Dakar, Senegal
[c] Infectious diseases department, Fann University Hospital Center, Dakar, Senegal
[d] Surgery department, Gaston Berger University, Saint-Louis, Senegal
[e] Radiology department, Gaston Berger University, Saint-Louis, Senegal

ARTICLE INFO

Keywords:
Fitz-Hugh-Curtis syndrome
Perihepatitis
HIV
Splenic tuberculosis
Ultrasound

ABSTRACT

Fitz-Hugh-Curtis (FHCS) is characterized by an inflammation of the hepatic capsule concomitant or following pelvic infection due to *Chlamydia trachomatis* or *Neisseria gonorrhea*. It is a rare condition occurring most often in a woman of childbearing age and very rare in male patients. Splenic involvement is also a rare form of abdominal tuberculosis. The association of these two conditions is very uncommon. We report the exceptional case of a 58-year-old HIV-positive male patient, with whom abdominal ultrasound helped diagnose FHCS associated with abdominal tuberculosis invovlving the spleen.

Introduction

Perihepatitis or Fitz-Hugh-Curtis syndrome (FHCS) is characterized by an inflammation of the liver capsule associated or following pelvic infection with *Neisseria gonorrhea or Chlamydia trachomatis* [1]. It is a rare condition occurring most often in a woman of childbearing age [2]. Cases reported in male patients remain rare and sporadic in the literature [3]. Its definitive diagnosis is based on laparotomy or laparoscopy showing violin string-like adhesions between the hepatic capsule and the abdominal wall [4]. In resources limited context, where laparoscopy is not always available, abdominal ultrasound could be used to evoke this diagnosis and retain it if other clinical and biological signs are in favor [1,5].

Abdominal tuberculosis occurs most often upon reactivation of pulmonary tuberculosis in immunocompromised patients. It is often the case particularly in HIV-positive patients. Splenic involvement is one of the rarest forms of abdominal tuberculosis and its definitive diagnosis often requires invasive means [6,7]. However, there are ultrasound signs for splenic involvement in tuberculosis which should be considered. Particularly, when the clinical context is suggestive and invasive diagnostic method are not available [8,9].

We report the case of a 58-year-old HIV-positive male patient with immunosuppression, in whom the ultrasound suggested the diagnosis of FHCS associated with splenic tuberculosis. The association of FHCS with tuberculosis is exceptional in the literature [4].

Case report

This is a 58-year-old male patient, married with 3 children, HIV-positive for 12 years. He complained of pain in the right upper quadrant of the abdomen evolving for the past five days associated with vespero-nocturnal fever with chills and sweating. There was no cough, nausea, vomiting or other digestive symptoms. The patient had been on triple antiretroviral therapy since being diagnosed with HIV and had voluntarily stopped it for 7 months without specific reason. In his history, there were no reported opportunistic infections. However, 3 days before his admission, there were two episodes of purulent urethral discharge associated with painful urination. He was sexually active only with his wife and denies high-risk sexual behavior. His last CD4 count 8 months earlier was 800 / mm3 with undetectable viral load. On admission the patient was in poor general condition, was pale and had a mild fever of 37.7 °C. The examination found tenderness of the right

* Corresponding author at: BP 5035 Fann, Dakar, Senegal.
E-mail address: niangibrahimaniang@gmail.com (I. Niang).
[1] Orcid: 0000-0002-6295-4518

https://doi.org/10.1016/j.idcr.2024.e02036
Received 13 September 2023; Received in revised form 21 July 2024; Accepted 29 July 2024
Available online 30 July 2024

hypochondrium without a palpable mass. The urethral discharge was no longer present and there were no other noticeable abnormalities. The biological exams showed a white blood cell count (WBC) of 18,000 elements / mm3 with 45 % lymphocytes, microcytic anemia (Hb 9 g / dl, MCV 72 fL), high C-reactive protein (CRP) (25 mg / dL) and erythrocyte sedimentation rate (ESR) (70 mm). His CD4 count was 250 / mm3 with a viral load of 16,000 copies / ml. Cytobacteriological examination of urine was positive for *Chlamydia trachomatis.* Liver and kidney function tests were normal. The chest x-ray was also normal.

Abdominal ultrasound revealed an anterior perihepatic fluid collection between the hepatic capsule and the diaphragm, containing fine pseudo-septa detected by the high-frequency probe (Fig. 1A-B). Ultrasound also found intra-abdominal lymphadenopathy (Fig. 2 A-B). The spleen was slightly increased in size with the presence of multiple microabscesses in the form of hypoechoic nodular lesions of millimeter-sizes diffused throughout the parenchyma and visualized with the high-frequency probe (Fig. 3 A-B). The liver and gallbladder appeared normal, as were the kidneys and other abdomino-pelvic organs. There was no peritoneal effusion. Considering previous episodes of purulent urethral discharge, perihepatic effusion on ultrasound, diffuse splenic micro-abscesses and abdominal lymphadenopathy in the context of HIV-positive, the diagnosis of FHCS associated with splenic tuberculosis was retained. He had antibiotic therapy with Doxycycline 100 mg x 2 per day during seven days and ceftriaxone 1 g in a single IV dose, in combination with anti-tuberculosis treatment (isoniazid, rifampicin, pyrazinamide and ethambutol). The patient had refused the scheduled reintroduction of his antiretroviral therapy. After 11 days of hospitalization, the abdominal pain subsided and there were no new episodes of urinary symptoms. A follow-up abdominal ultrasound revealed regression of the perihepatic collection without any other new anomaly. But the patient's general condition remained altered and the patient died on the 12th day of hospitalization. His family objected to performing an autopsy.

Discussion

FHCS is characterized by an inflammation of the hepatic capsule concomitant with or following pelvic infection with *Chlamydia trachomatis* or *Neisseria gonorrhea* [1]. It most often occurs in sexually active women with high-risk sexual behavior [10]. It is rare or even exceptional in male patients [3]. The pathophysiological mechanism could be at the origin of this difference in prevalence. In women, FHCS is thought to result from bacterial migration from the pelvis to the hepatic capsule via the parietocolic gutter, while in men perihepatic contamination occurs via the hematogenous or lymphatic spread [2]. As in our patient, pain in the right upper quadrant is the most common symptom [2]. But this sign is not very specific and may lead to considering several diagnoses including cholecystitis, liver abscess, subphrenic abscess … *etc*. These conditions are more frequent and could be the cause of delayed diagnosis or misdiagnosis of FHCS which is rarer. Thus, in both women and men, the concomitant presence of uro-genital signs of infection (urethral discharge, painful urination,) and upper quadrant pain should lead evoking this diagnosis to carry out appropriate explorations. The germs involved (*Neisseria gonorrhea and Chlamydia trachomatis*) should be looked for in the urine or on a genital swab. PCR test has a greater sensitivity than direct examinations and cultures [11].

The diagnosis of fitz hugh curtis disease in male patients is ideally made by laparoscopy which shows the appearance of violin strings [12]. When laparoscopy is unavailable, the diagnosis can be based on clinical, urinary and blood tests that allow to eliminate differential diagnoses as well as imaging (Ultrasound and CT) [13].

In our patient, cytobacteriological examination of urine was positive for *Chlamydia trachomatis*, which confirmed the diagnosis of FHCS. The additional argument in our patient was the ultrasound and the perihepatic effusion associated with the pseudo-septas. This ultrasound sign has been described in other reported cases [14,15]. The pseudo-septas correspond to the "violins strings" found in laparotomy and laparoscopy and corresponding to adhesions between the hepatic capsule and the abdominal wall or diaphragm. Laparoscopy is the best diagnostic tests (showing violins strings-like adhesions). However, it is not always feasible due to lack of resources or problems related to the patient [5, 16]. Thus, imaging constitutes an alternative non-invasive diagnostic tool. The CT signs suggestive of the diagnosis of FHCS are more described and in the form of linear uptake of the liver capsule in the early phase, associated or not with perihepatic effusion [17]. Ultrasound signs are rarely reported and are limited to perihepatic fluid effusion [15]. Because of its greater accessibility in resource limited settings, ultrasound should be reconsidered. In fact, it can easily eliminate other more usual differential diagnoses. Splenic tuberculosis is a rare condition in abdominal tuberculosis. Its prevalence has increased with the pandemic of HIV-TB co-infection particularly in developing countries [18]. Its diagnosis is most often made with abdominal ultrasound, which usually finds micro abscesses in the form of multiple focal hypoechoic

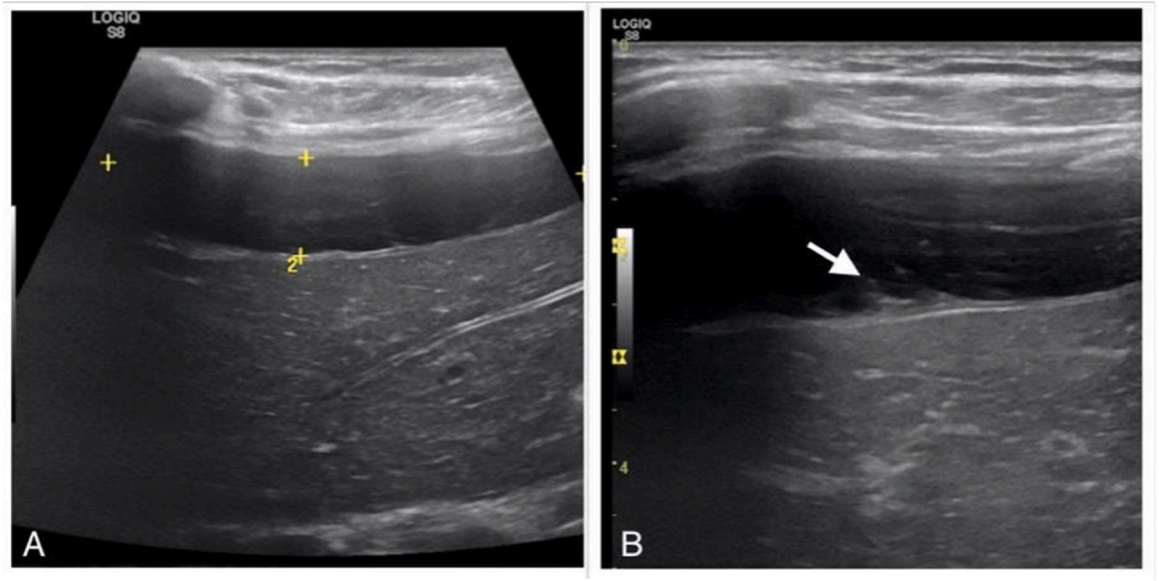

Fig. 1. perihepatic free fluid collection. A) with the low-frequency probe, perihepatic collection between the hepatic capsule and the anterior abdominal wall. B) with the high-frequency probe, visualization of the pseudo-septa corresponding to the "violin string".

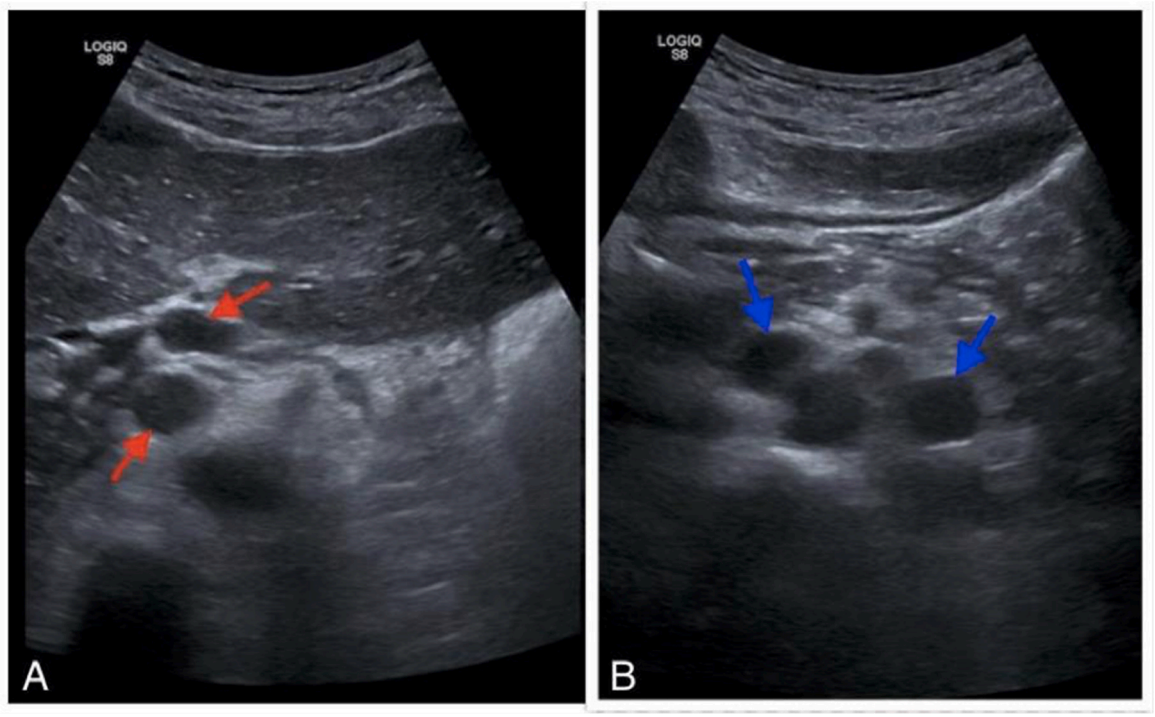

Fig. 2. intraabdominal lymphadenopathies. A) lymphadenopathies of the hepatic hilum, almost in contact with the liver (red arrows). B) coeliomesenteric lymphadenopathy, opposite the celiac trunk and mesenteric artery (blue arrows).

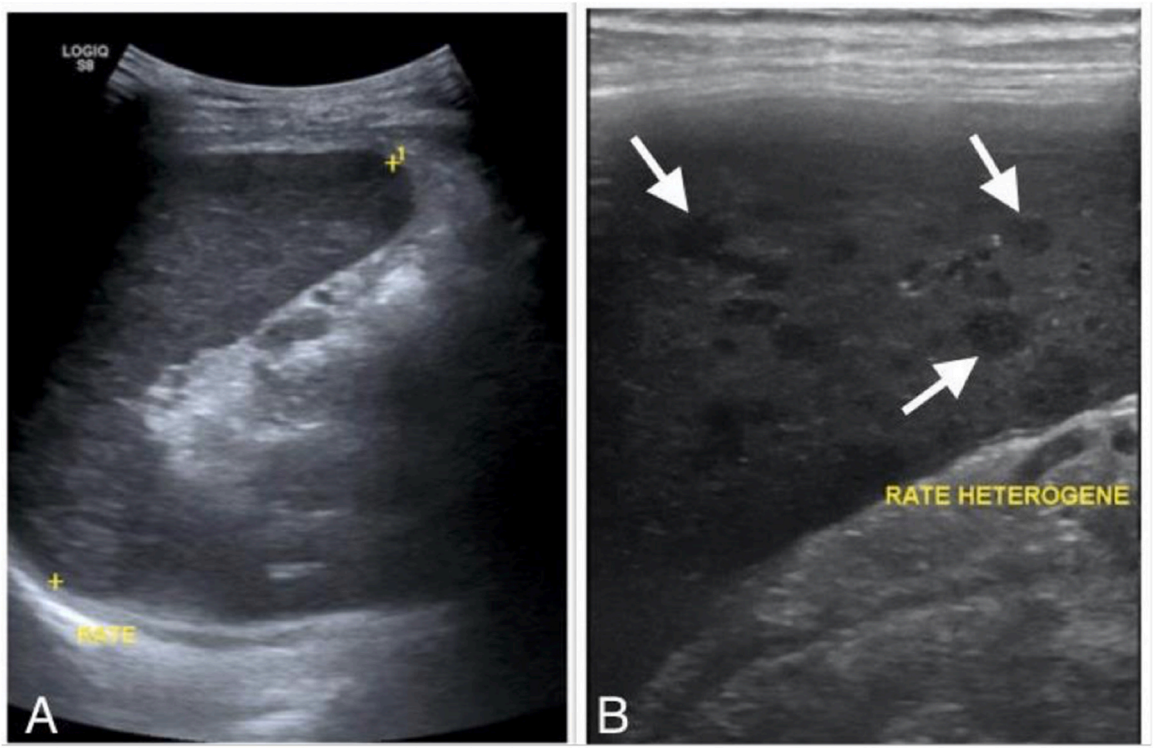

Fig. 3. spleen. A) Sagittal view with the low-frequency probe: discrete splenomegaly B) exploration with the high frequency probe making it possible to visualize multiple hypoechoic lesions of millimeter sizes (white arrows), corresponding to the mircoabcesses.

lesions, corresponding to granulomas [9,18]. Ultrasound can also look for intra-abdominal lymphadenopathy or other intra-abdominal tuberculosis localizations that may be associated. The diagnosis of tuberculosis is usually made by sputum examination, culture, biopsy, or molecular detection method [19]. Diagnosis of extrapulmonary tuberculosis in patients with HIV infection, however, is difficult in the absence of active pulmonary tuberculosis. In the presence of HIV, sputum examinations or cultures to diagnose extrapulmonary tuberculosis are unreliable and are only positive in only 3 % of patients [20]. Regarding splenic involvement, diagnostic confirmation is made by

pathological examination after fine-needle aspiration, biopsy or on a splenectomy specimen [21]. One study reported the possibility of using ultrasound with contrast to help differentiate between splenic tuberculosis and splenic lymphoma [22].

For some authors in areas with high tuberculosis endemicity, the presence of splenic micro abscesses on ultrasound is sufficiently suggestive of splenic tuberculosis to make the diagnosis and start antituberculosis treatment [9,23]. This is the attitude we chose in our patients because our resources did not allow us to perform a fine-needle aspiration and a biopsy would be too invasive. Ultrasound thus allows more efficient care due to its low cost and accessibility. In addition, it allows follow-up after treatment.

The association of these two conditions in our patient, although coincidental is exceptionally rare in the literature [4]. In our patient, the existence of immunodepression and refusal to take antiretroviral treatment for more than 7 months could be the cause of the fatal course.

In conclusion, the diagnosis of FHCS and splenic tuberculosis in an area with limited resources is difficult. Abdominal ultrasound plays a key role and its resultsshouldbeconsidered to overcome the inaccessibility of invasive diagnostic tools.

Funding

None.

CRediT authorship contribution statement

Abdoulaye Dione Diop: Writing – review & editing. **Sokhna Ba:** Writing – review & editing. **Ibrahima NIANG:** Validation, Writing – original draft, Writing – review & editing, Conceptualization. **Daouda Thioub:** Investigation, Writing – review & editing. **Mamadou Ly:** Investigation, Writing – review & editing. **Abdourahmane Ndong:** Writing – review & editing. **Fallou Galass Niang:** Writing – review & editing.

Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

Acknowledgments

None.

Author agreement

The patient's wife signed a free and informed consent to the anonymous.

publication of the material contained in this article.

Patient consent

The patient's wife has signed an informed consent form.

References

[1] Lisičar I, Begovac J, Zekan Š. A rare case of Fitz-Hugh-Curtis syndrome caused by Chlamydia trachomatis in an HIV-positive male patient. SAGE Open Med Case Rep 2019;7. 2050313X18823592.
[2] Woo SY, Kim JI, Cheung DY, Cho SH, Park SH, Han JY, et al. Clinical outcome of Fitz-Hugh-Curtis syndrome mimicking acute biliary disease. World J Gastroenterol 2008;14(45):6975–80.
[3] Nardini P, Compri M, Marangoni A, D'Antuono A, Bellavista S, Calvanese C, et al. Acute Fitz-Hugh-Curtis syndrome in a man due to gonococcal infection. J Emerg Med 2015;48(3):e59–62.
[4] Coremans L, de Clerck F. Fitz-Hugh-Curtis syndrome associated with tuberculous salpingitis and peritonitis: a case presentation and review of literature. BMC Gastroenterol 20 mars 2018;18(1):42.
[5] Di Rocco G, Giannotti D, Collalti M, Mele R, Pontone S, Frezzotti F, et al. Acute abdominal pain in a 24-year-old woman: Fitz-Hugh-Curtis syndrome associated with pyelonephritis. Clin Sao Paulo Braz déc 2012;67(12):1493–5.
[6] Rodarte-Shade M, Diaz-Elizondo JA. Splenic tuberculosis. Surg Infect déc 2012;13 (6):420–1.
[7] Guidi R, Bolli V, Lanza C, Biagetti C, Osimani P, de Benedictis FM. Macronodular hepatosplenic tuberculosis. Acta Radio Short Rep 2012;1(5). arsr.2012.12-0020.
[8] Malik A, Saxena NC. Ultrasound in abdominal tuberculosis. Abdom Imaging 2003; 28(4):574–9.
[9] Schafer JM, Welwarth J, Novack V, Balk D, Beals T, Naraghi L, et al. Detection of splenic microabscesses with ultrasound as a marker for extrapulmonary tuberculosis in patients with HIV: a systematic review. South Afr Med J Suid-Afr Tydskr Vir Geneeskd 26 juill 2019;109(8):570–6.
[10] Rivero-Sánchez L, López-Soriano EM, Guarner-Aguilar L. Fitz-Hugh-Curtis syndrome: abdominal pain in women of 26 years old. Rev Esp Enferm Dig oct 2011; 103(10):546–8.
[11] Workowski K.A., Bolan G.A., Centers for Disease Control and Prevention. Sexually transmitted diseases treatment guidelines, 2015. MMWR Recomm Rep Morb Mortal Wkly Rep Recomm Rep. 5 juin 2015;64(RR-03):1-137.
[12] Rouhard S, Maldague P, Ramboux A. Fitz-Hugh-Curtis syndrome in a man. Endoscopy 2014;46(Suppl 1). UCTN:E1.
[13] Yi H, Shim CS, Kim GW, Kim JS, Choi IZ. Case of Fitz-Hugh-Curtis syndrome in male without presentation of sexually transmitted disease. World J Clin Cases 2015;3(11):965–9.
[14] Banerjee B, Rennison A, Boyes BE. Sonographic features in a case of Fitz-Hugh-Curtis syndrome masquerading as malignancy. Br J Radio 1992;65(772):342–4.
[15] van Dongen PW. Diagnosis of Fitz-Hugh-Curtis syndrome by ultrasound. Eur J Obstet Gynecol Reprod Biol juill 1993;50(2):159–62.
[16] Ricci P, Lema R, Solá V, Fernández C, Fabres C, Fernández E, et al. Fitz-Hugh-Curtis syndrome: three cases of incidental diagnosis during laparoscopy. J Obstet Gynaecol J Inst Obstet Gynaecol avr 2008;28(3):352–4.
[17] Nishie A, Yoshimitsu K, Irie H, Yoshitake T, Aibe H, Tajima T, et al. Fitz-Hugh-Curtis syndrome. Radiologic manifestation. J Comput Assist Tomogr 2003;27(5): 786–91.
[18] Sharma SK, Mohan A. Extrapulmonary tuberculosis. Indian J Med Res oct 2004; 120(4):316–53.
[19] Kwanjana IH, Harries AD, Hargreaves NJ, Van Gorkom J, Ringdal T, Salaniponi FM. Sputum-smear examination in patients with extrapulmonary tuberculosis in Malawi. Trans R Soc Trop Med Hyg 2000;94(4):395–8.
[20] Harries AD, Maher D, Nunn P. An approach to the problems of diagnosing and treating adult smear-negative pulmonary tuberculosis in high-HIV-prevalence settings in sub-Saharan Africa. Bull World Health Organ 1998;76(6):651–62.
[21] Hamizah R, Rohana AG, Anwar SA, Ong TZ, Hamzaini AH, Zulkarnaen AN. Splenic tuberculosis presenting as pyrexia of unknown origin. Med J Malays 2007;62(1): 70–1.
[22] Zhang W, Yang G, Zhang X, Ni T. The role of contrast-enhanced ultrasound in differentiating splenic tuberculosis from Splenic Lymphoma. Front Oncol [Internet] 2022:12 [cité 4 juill 2024].
[23] Heller T, Wallrauch C, Goblirsch S, Brunetti E. Focused assessment with sonography for HIV-associated tuberculosis (FASH): a short protocol and a pictorial review. Crit Ultrasound J 2012;4(1):21.