

Tubo-Ovarian Mass with raised CA-125 in 21-year-old female

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Case Report

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Abstract

Introduction: Peritonitis associated with fungal species *Curvularia lunata* seldom occurs with only five cases reported in literature, all in middle age patients with comorbidities undergoing dialysis.

Case Report: A 21-year-old female who was referred to surgical oncology OPD with a diagnosis of ovarian malignancy, based on raised **cancer antigen 125** (CA 125) and suspected tubo-ovarian mass (TOM) on MRI. Review of the MRI showed pelvic collection with TOM, suggestive of infective pathology. Ultrasound guided aspiration of the cystic collection identified the presence of *Curvularia lunata*. She was treated with oral Itraconazole which showed symptomatic improvement and radiological response. In the follow-up period the patient developed chest wall swelling, aspiration and geneXpert® revealed MDR tuberculosis and treatment was started.

Conclusions: Unusual causes of TOM and raised CA 125 should be kept in mind when dealing with young patients, as the possibility of epithelial ovarian cancer in this age is very low.

Introduction

Fungal peritonitis is an uncommon entity, which is associated with significant morbidity and mortality. Although, most of the fungal peritonitis cases that have been reported are caused by *Candida albicans*, there are other rare fungal species that has also been identified¹. *Curvularia lunata* is one such organism which seldom causes infection in human. It is a saprobic dematiaceous fungus that is primarily found in the soil².

C. lunata was first described in 1898 by Wakker and Went, when they isolated it from dead sugar cane leaves. It was first suspected as human pathogen in 1959, when Baylet isolated it from black grain mycetoma in Senegal². Review of literature revealed a handful of cases caused by *C. lunata* in human, which includes mycetoma, paranasal sinusitis, peritonitis, cerebral phaeohyphomycosis and disseminated disease infecting the eye, skin, pleura, lung, spine, brain etc. In this article, we report a rare case of fungal peritonitis associated with *Curvularia lunata* in a female followed by multidrug resistant (MDR) tuberculosis.

Case

A 21-year-old sexually virgin female was referred to Surgical Oncology out patient in June 2020 with provisional diagnosis of epithelial ovarian malignancy on the basis of MRI findings and raised CA125 level of 129.8 U/ml (normal 0–35). On enquiring, she had irregular menstruation for last 6 months, abdominal distension for 2 months and pain in lower abdomen for last 1 month. She had a past history of cervical lymph node tuberculosis in 2017, for which she completed anti tubercular therapy for 18 months with standard regimen of isoniazid, rifampicin, pyrazinamide, and ethambutol. Other than this there was no significant medical and surgical history. Personal and family history was noncontributory. On clinical examination, general condition was fair, vitals were stable, systemic examination was normal.

On per abdomen examination, patient had mild tenderness in the lower abdomen with no signs of guarding, rigidity or free fluid in the abdomen. No definite mass or lump was palpable.

Her previous MRI scan was reviewed which showed extensive smooth peritoneal thickening with moderate ascites, multiloculated cystic lesion in bilateral adnexa, variably hyperintense on T2 weighted MRI and variably hyperintense to hypointense on T1 weighted images, the lesions were in close relation with ovaries but bilateral ovaries were normally visualized (Fig. 1). This suggested possibility of tubo-ovarian origin of the mass. Her routine biochemical blood investigations were within normal limit except for raised ESR 32mm/hr (normal 0–20). Other tumor markers were also within normal limit, LDH- 216 U/L (normal 135–225), alpha fetoprotein-1.6 ng/ml (normal 0.89–8.78), serum CEA < 0.5 ng/ml (normal 0–5), beta HCG < 2 mIU/ml (normal 0–5). Seeing her past history of tuberculosis, interferon gamma release assay was done which was negative. After this primary diagnosis of tubo-ovarian abscess was made.

A transabdominal USG guided aspiration of the fluid done and sent for geneXpert®, cell cytology, cell block and culture (bacterial/fungal/tubercular). GeneXpert® was negative for tuberculosis. The fluid cytology was negative for malignant cells and the cell block showed reactive mesothelial cells and neutrophils. The microbiological reports revealed a sterile bacterial culture. On fungal culture, fungal elements were grown that on Matrix assisted laser desorption ionization Time of flight – Mass spectrometry (MALDI-TOF MS) (Bruker Daltonics, Germany), showed it to be *Curvularia lunata*

She was started on oral standard Itraconazole 200mg once daily and was followed-up every week. During the second week of follow-up, an abdominal CECT was repeated that showed bilateral adnexal masses with dilated tubular structures with thickened enhancing walls, moderate fluid collection was still seen in the pelvis along with peritoneal thickening, and the lesion also showed calcifications and cystic components of various compositions suggesting diagnosis of a tubo ovarian abscess (Fig. 2). So, another therapeutic USG guided aspiration was done and 80 ml fluid was aspirated.

After 6 weeks of Itraconazole treatment, she was symptomatically improved Repeat MRI pelvis was done, that showed near total resolution of mass, bulk of bilateral ovaries was seen normally. Figure 2). Again, transabdominal ultrasound guided fluid aspiration was done from remaining cyst and sent for repeat fungal culture which was negative for any fungal elements. Itraconazole was stopped and she was kept on regular follow up. Two months later patient presented with a chest wall swelling and weight loss. A CT scan was done that showed miliary tuberculosis and cold abscess in the chest wall (Fig. 3). An aspiration was done and sent for geneXpert® that confirmed MDR tuberculosis, 3 months after the start of antitubercular treatment the cold abscess was regressed and the patient had reported 3 Kg of weight gain.

Discussion

Fungal peritonitis associated with *Curvularia lunata* is a relatively rare phenomenon. To the best of our knowledge, a total of ten cases of peritonitis caused by the fungal species of *Curvularia* (see

supplementary material, table 1) has been reported in English literature³⁻¹² and half of these cases are due to *C. lunata*. All *Curvularia* infections which are reported have been seen only in immunocompromised patients with type 2 Diabetes, hypertension, ischemic heart disease and end stage renal disease. In this patient there was a previous history of tuberculosis for which she took ATT and this could be a reason for her compromised immunity. Because of this she acquired *C. lunata* infection as an opportunistic infection in presence of her decreased immunity¹³ and again she probably had a re-emergence of her latent TB infection, because of her decreased immunity¹⁴. Tiemesson et al mentioned that ATT intake can cause defective neutrophil function and affect innate immunity¹⁵.

Curvularia usually spread via inhalational or dermal inoculation routes. All cases which are reported have dermal inoculation route through peritoneal dialysis catheter. Three forms of *Curvularia* infection have been observed. They are catheter obstruction without peritonitis, catheter obstruction with peritonitis and only peritonitis. The present case falls under the last category i.e. only peritonitis.

TOA are usually a consequence of pelvic inflammatory disease (PID) and most commonly seen in sexually active women of reproductive age. Cho *et al.*, reported 4.1% incidence of virginal women PID among all cases of PID. Several hypothesis have been proposed for origin in virgin women through lower genital tract, urinary tract, gastrointestinal tract and skin wound but none of them were proven¹⁶. Early diagnosis is the key for management and ultrasound guided aspiration and antibiotics is the valid first line treatment option as we have done in our case. Surgical exploration only needed in acute cases, if ruptured TOA is suspected¹⁷.

Differentiating these masses on imaging from bacterial or tubercular infection is also important. In genitourinary infections, fungal infections on CT range from homogeneous hypo attenuating masses to heterogeneous masses with areas of hypo enhancement or calcifications. On MR images, the appearance varies according to the acuity of the infection, extent of necrosis, and degree of calcification. Most frequently, the masses are T1 hypointense and mildly T2 hyperintense¹⁸. Differential diagnosis of abdominal tuberculosis also cannot be ruled out as in wet type it presents primarily either as free or loculated ascites, associated or not with diffuse and smooth peritoneal thickening; in dry type of presentation there is predominance of peritoneal and mesenteric thickening with caseous nodules, lymph nodes enlargement and fibrinous adhesions; and in the fibrous type of presentation is characterized by omental thickening, entanglement of bowel loops, loculated ascites¹⁹.

In young women, most cases of TOA mimics as pelvic tumours even after imaging studies and they usually undergo surgical procedure which can easily be avoided and treated by appropriate medical management as the first line of treatment. A radiologist experienced with pelvic imaging is necessary for making or excluding the diagnosis before deciding any plan of management. Of all the *C. lunata* peritonitis cases reported, this is the first case that presented with TOA with peritonitis in virgin woman.

Declarations

Conflict of Interests

Authors declare that there are no conflicts of interests

Ethics and consent

Written informed consent was obtained from patient for publication of this case and accompanying images

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Consent to publish

The consent to publish the case report and accompanying images was obtained from the patient.

Authors Contribution

Roli Purwar, Literature search, preparation of the manuscript, overall coordination

Kishan Soni, Literature search, preparation of the manuscript

Ragini Tilak, Preparation of the manuscript related to microbiological part, diagnosis and editing of the manuscript

Ashish Verma, Preparation of Radiological part of manuscript, images and description, editing of the manuscript

Manoj Pandey: Concept and design, final editing

All authors have read and approved the manuscript

Availability of data

This is a case report the data is provided in the text of the manuscript.

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Figures

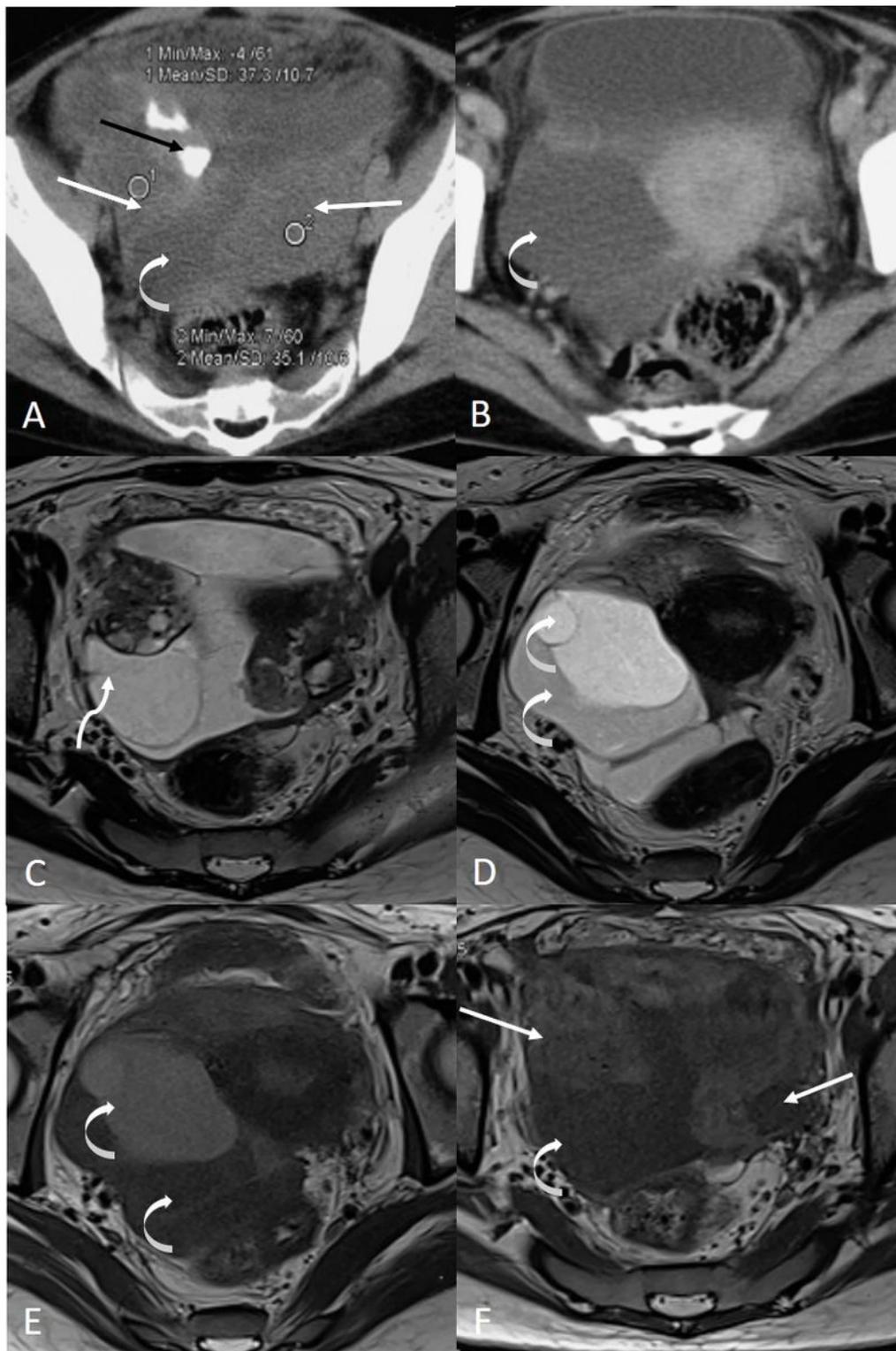


Figure 1

Axial MRI images of pelvis before treatment showing uterus in the centre with a multiloculated cystic lesion being hypoattenuating on CT scan (A,B), variably hyperintense on T2 weighted MRI (C,D) and variably hyperintense to hypointense on T1 weighted (E,F) images. The lesion also shows calcification on CT scan (black straight arrow in A) and variable intensity pattern (curved arrows) suggesting that there are cystic components of various compositions. The lesion also shows a partial septa sign (twisted arrow

in C), this along with close circumferential relation with ovaries (straight white arrows in all sections) clearly indicated towards a possible tubo-ovarian origin of the mass. The bulk of both ovaries is however visualized normally (straight white arrows).

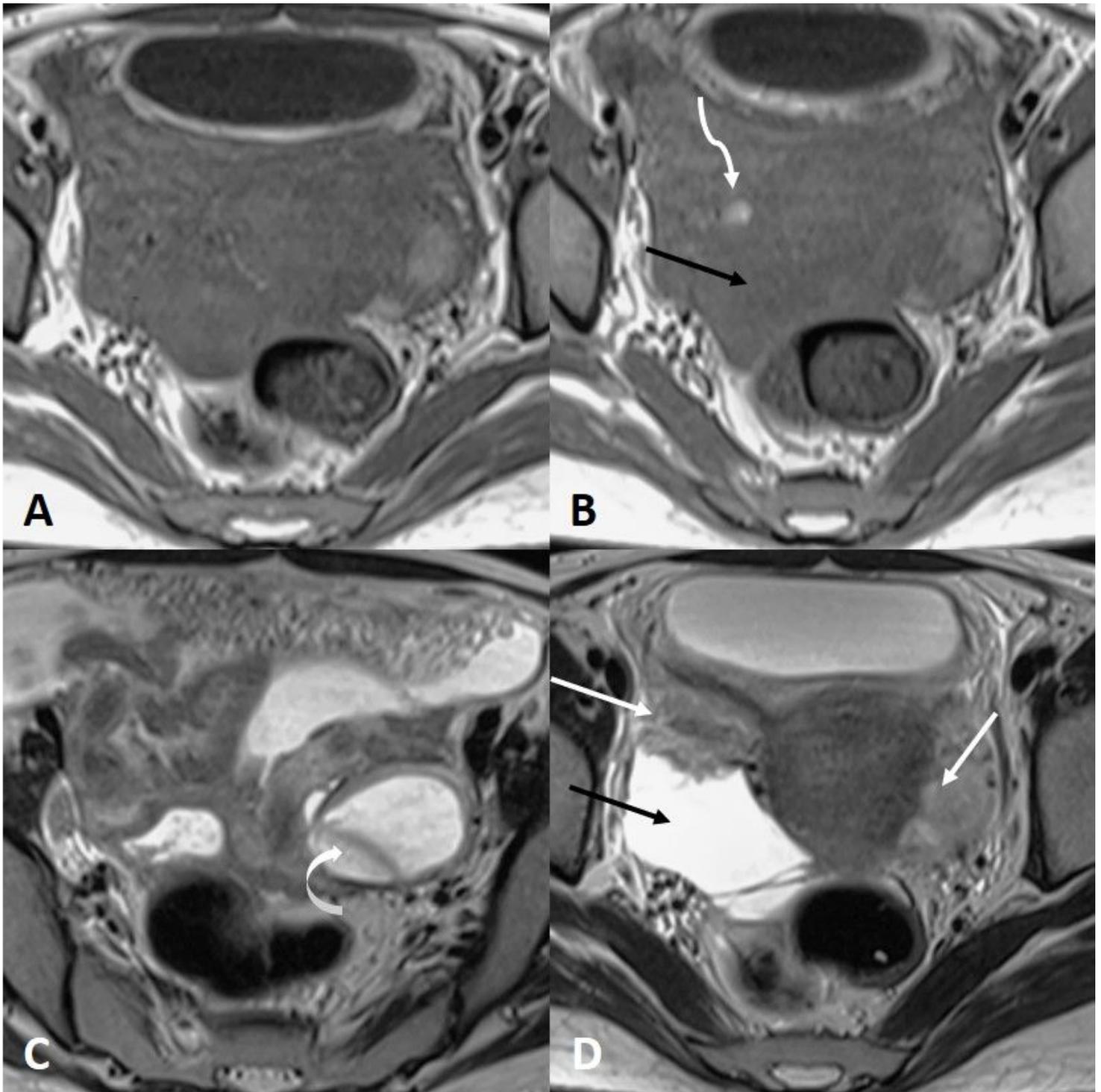


Figure 2

Axial MRI images of pelvis after treatment showing uterus in the centre with near total resolution of previously seen mass. Only a small component which has verted to the left side (curved arrow) is

remaining. The previously seen calcification is still noted (twisted arrow) The bulk of both ovaries is however visualized normally (straight white arrow). Mild ascites is seen (straight black arrow) which may indicate post-treatment residual peritumoral inflammation.

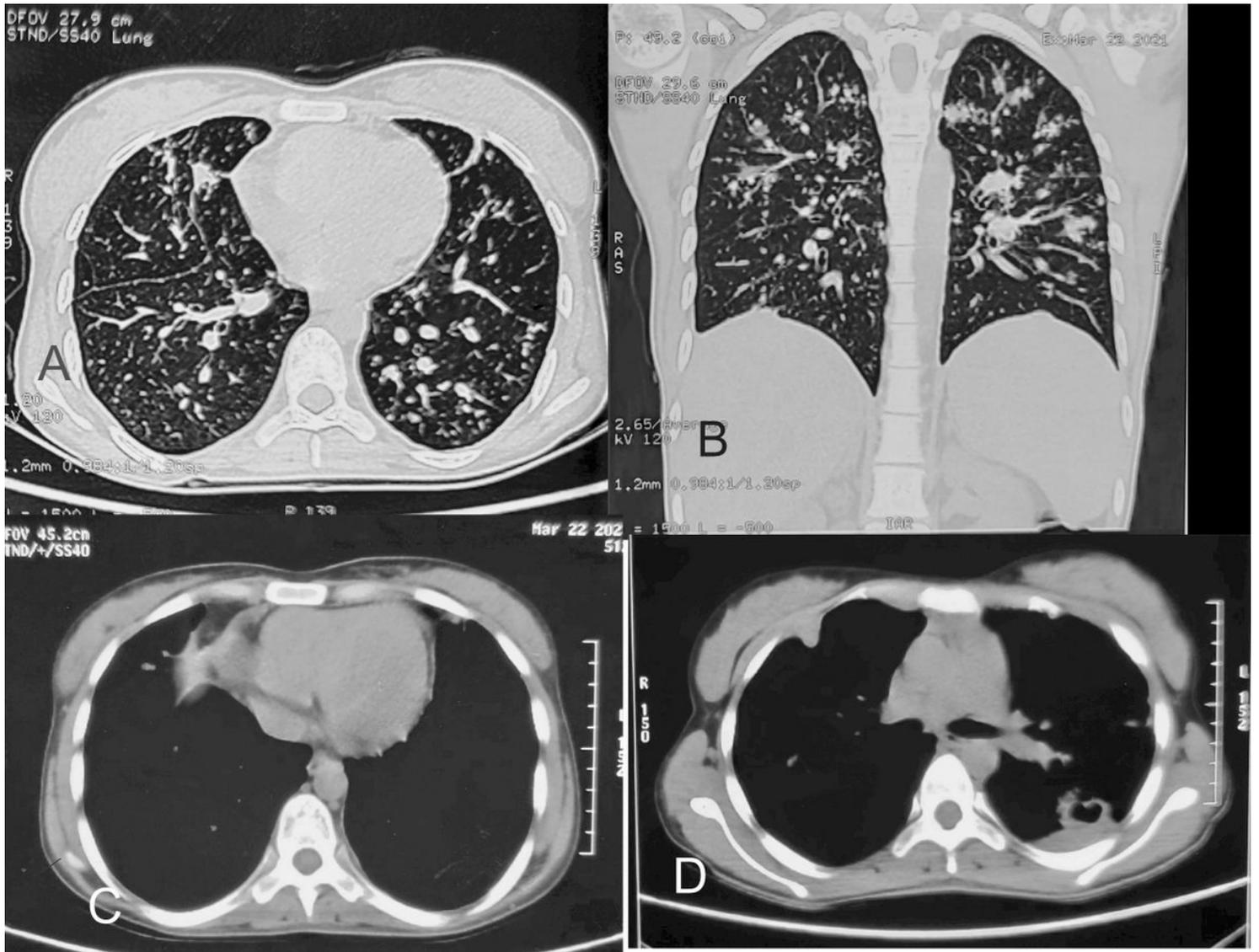


Figure 3

Computerized tomography scan of the lung and thorax showing A) Axial section of bilateral lung showing disseminated tuberculosis lesions in both lungs, B) sagittal section showing the parenchymal lesions C) Axial section showing sub mammary plural collection and D) axial section showing subcutaneous collection (cold abscess) at 7 O clock position