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Case report

Pericardial effusion and mass : a rare presentation of Chronic Myelogenous Leukemia.

Arti Muley[*a], Mahima Chajer[b], Hetal Pandya[b], J D Lakhani [d], Chaitri Shah [e], Ashwin Patel [f]

[a] *Asstt Prof Medicine, [b]Assoc. Prof Medicine, cCardiologist , [d]Prof. &HOD, Medicine, eIntensivist, Medicine, fResident, Medicine, SBKS Medical College and Research Institute, SumandeepVidyapeeth.Pipariya,Vadodara,Gujarat. India.

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ABSTRACT

Chronic myelogenous leukemia (CML) mostly presents as anemia, fatigue and breathlessness with hepatosplenomegaly. Pericardial involvement with effusion has been reported in acute leukemia but very rarely in CML. Only four reports of CML presenting with pericardial effusion are available till now. There is only one report of presentation with atrial mass. We present a case who presented with CML associated pericardial effusion and mass which resolved with chemotherapy with hydroxyurea. The finding becomes more important as there are recent reports of pericardial effusion being seen as adverse effect of the drugs imatinib and dasatinib. This case presented with pericardial effusion prior to starting the chemotherapy and resolved on treatment. It brings to notice a need for more research to find out the status of pericardial involvement in cases of CML and to rule out presence of pericardial effusion prior to starting treatment with imatinib and dasatinib.

1. Introduction

Chronic myelogenous leukemia (CML) mostly presents as anemia, fatigue and breathlessness with hepatosplenomegaly. Pericardial involvement with effusion has been reported in acute leukemia but very rarely in CML. As per our knowledge, only four reports of CML presenting as pericardial effusion are available till now [1-4]. We present a case who presented with CML associated pericardial effusion and mass which resolved with chemotherapy. The finding becomes more important as there are recent reports of pericardial effusion being seen as adverse effect of the newer drugs imatinib and dasatinib. Since our case presented with pericardial effusion even prior to starting the chemotherapy and resolved on treatment, it brings to notice a need to do 2 D Echo in all cases of CML to rule out pericardial effusion before starting these drugs.

2.Findings:

In May 2011, a 25-year-old female was admitted to our hospital with low grade fever without chills since 1 month, dry cough since 20 days , progressive dyspnea since 15 days and chest pain (dull aching, localised and aggravated with lifting weight) since 3 days. Appetite was reduced. Menstrual history was normal. Physical examination revealed mild pallor .Vitals were normal with pulses normally felt in all four limbs. Systemic examination revealed muffled heart sounds , pericardial rub and mildly enlarged spleen. There were no skin lesions.

Routine blood investigations showed Hemoglobin 10.1 g/dL, WBC 43,600/micro L (79% neutrophils,12%lymphocytes) and platelets 6,90,000/microL. Renal function test, liver function test and urine were normal. CXR PA showed enlarged cardiac silhouette. USG abdomen showed mild hepato-splenomegaly with pericardial effusion. HIV & HBsAg were negative. On 2 D-echocardiography, chambers were normal with mild tricuspid regurgitation and moderate pericardial effusion. Along with the effusion, an echogenic mass was also visible in the right AV groove (see Fig1). CT thorax showed moderate pericardial effusion with thrombosis in leftbrachiocephalic vein. The mass was not visible in contrast enhanced CT or MRI.

Blood culture was sent, pericardial fluid tapping was planned and broad spectrum antibiotics were started empirically with other symptomatic and supportive treatment. Despite treatment, fever persisted and no improvement was seen in the TLC. Diagnostic tapping of pericardial fluid was done. As the mass was

* Corresponding Author : Dr Arti Muley,
Asstt Prof Medicine,
SBKS Medical College and Research Institute,
SumandeepVidyapeeth.Pipariya,
Vadodara,Gujarat. India.
E-mail: muleyarti@yahoo.com

not approachable and biopsy from mass was not possible (due to risks involved) , pericardial biopsy was done. Pericardial fluid was hemorrhagic and was reported to be having pH7.5, proteins 5.6,sugar132, ADA 81, TLC 50,000 (lymphocytes 14%, polymorphs 48%, mononucleated cells 36% and eosinophils 2%). Gram and Ziehl-Nielsen stains and cultures on pericardial fluid were negative. Blood culture and PPD test were also negative. Pericardial biopsy showed only inflammatory changes. In view of exudative pericardial effusion, raised ADA and high prevalence of tuberculosis in our country, anti tubercular treatment was started.

With appearance showing large areas of haemorrhage and necrosis.

Despite the treatment , symptoms did not improve and TLC repeated on sixth day of admission reached to 92,000/microL. Peripheral smear revealed 30% polymorphs, 18% lymphocytes, 20% myelocytes, 20% metamyelocytes, 10% band cells and 2% promyelocytes.

Bone marrow examination was suggestive of CML (35% metamyelocytes, 25% myelocytes, 10% promyelocytes, 10% band cells, 10% neutrophils, 1% blast cells, 5% cells of erythroid series and 4% lymphocytes. Myeloid to erythroid ratio was 20:1). Qualitative study for presence of Philadelphia chromosome was positive.

The diagnosis of CML with pericardial mass with pericardial effusion with left brachiocephalic vein thrombosis was established. Hydroxyurea was started (in view of recent reports about pericardial effusion being seen as adverse effect of the newer drugs e.g; imatinib and desatinib). ATT was stopped. Patient improved symptomatically, fever and breathlessness subsided, TLC started reducing and she was discharged on hydroxyurea. On her follow up visit after 15 days, 2D Echo was repeated which showed no pericardial effusion and the mass had also resolved.

3.Discussion

The most common cause of pericardial effusion in developing world is tuberculosis (especially in countries with high prevalence of tuberculosis) and raised ADA is considered almost diagnostic of tuberculosis in these countries. The reported cut off value for ADA varies from 47 to 60 U/L [8].

High ADA has also been reported in noninfectious conditions associated with pleural fluid lymphocytosis, including malignant conditions (eg, adenocarcinomas, acute leukemias, and lymphomas) and collagen vascular diseases (eg, rheumatoid pleuritis and systemic lupus erythematosus). In our case, the pericardial effusion had high ADA , but it finally turned out to be due to CML. This case highlights the need to completely investigate and rule out other causes of pericardial effusion as higher rate of false-positive results can lead to unnecessary administration of antitubercular therapy or a delay in making an alternative diagnosis.

The cause of pericardial effusion in CML may be leukemic infiltration, extramedullary hematopoiesis , infections and bleeding in CML. Looking at negative cultures, negative tests for tuberculosis, non enhancing nature of the mass, and resolution of mass with treatment, in our case, there was a strong possibility of extramedullary hematopoiesis although it could not be proved as biopsy from the mass could not be done..

We are discussing this case because the most frequent presentation in CML is with left upper quadrant fullness, decreased exercise tolerance and hepato-splenomegaly. Pericardial effusion as presentation in a case of CML has been described in only few case reports [1-4] previously. There is only one report of cardiac mass in a case of CML prior to this, which was an intra atrial mass. To our knowledge, there has been no report of pericardial mass in a case of CML before. This may be because of the fact that 2D Echo is not done routinely in patients of CML leaving a possibility of mild pericardial effusion remaining undiagnosed. We also emphasise on the fact that there is much research going on at present , on the new drugs for CML ; imatinib and desatinib which have shown that pericardial effusion is being seen very frequently after starting these drugs[5-7].This case brings to notice the need of 2D echo prior to starting with the newer drugs for CML to rule out pre-existent effusion.

Fig 1:Pericardial mass in right atrial groove with pericardial effusion on initial presentation, before starting hydroxyurea.

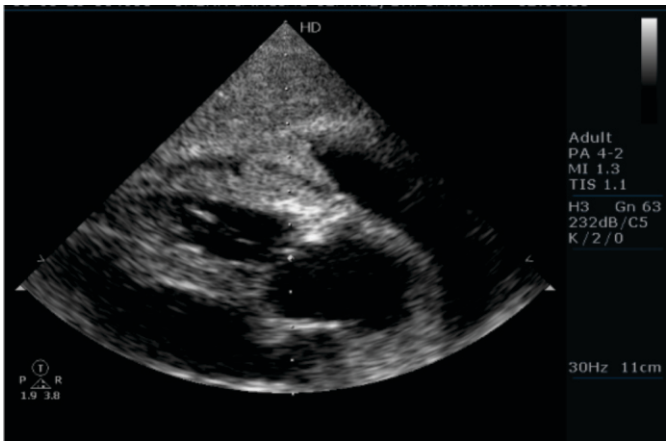
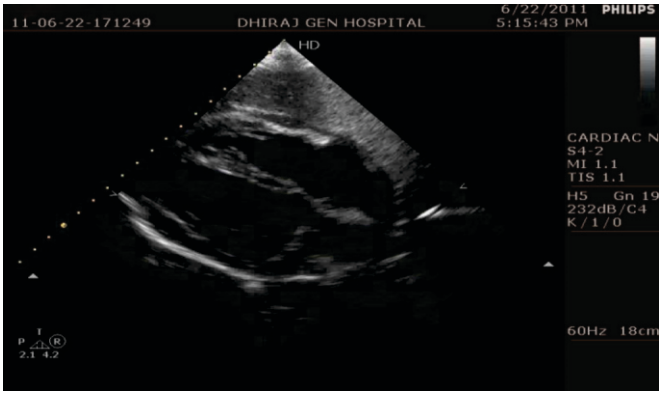


Fig 2: 2D Echo of the same patient 15 days after starting hydroxyurea.



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