Case Report

Received on 20.07.2016, **Accepted on** 03.08.2016

Indian Journal of Emergency Medicine
Volume 2 Number 2, July - December 2016
DOI: http://dx.doi.org/10.21088/ijem.2395.311X.2216.9

Case of New Onset Bochdalek's Hernia in Adult: Is it Thalidomide Induced?

Shivam Gupta*, Amit Kumar Jain*, Bidita Khandelwal, Parvati Nandy*****

Author's Affiliation:
*Resident, **Professor and HOD, ***Professor, Department of Internal Medicine, Sikkim Manipal Institute of Medical Sciences, Sikkim Manipal University, Tadong Sikkim-737102 Gangtok.

Corresponding Author:
B. Khandelwal, Professor and HOD, Department of Internal Medicine, Sikkim Manipal Institute of Medical Sciences Tadong Sikkim-737102 Gangtok.
E-mail: drbidita@gmail.com

Abstract

Bochdalek hernia is the commonest type of diaphgramatic hernia. It was first described in 1754 by Mc Culley. In 1848 Bochdalek described that diaphragmatic hernia occurs due to improper fusion of posterolateral foramina of diaphragm. We present a case of 48 year old female who first presented with chief complaints of epistaxis with cervical and lumbar back pain. She was diagnosed as a case of multiple myeloma and started on drug thalidomide. On follow ups the patient started having respiratory complaints for which she underwent X-ray chest and CT thorax which further revealed that patient had developed a new onset bochdaleks hernia as X-ray chest on her previous visits was normal. With no other preceding risk factors or previous respiratory complaints, we came to a conclusion that this could be a case where thalidomide, which till now has only experimentally proven to cause hernia, may be a cause of this new onset bochdaleks hernia.

Keywords: Bochdalek hernia; Thalidomide.

Introduction

Bochdalek hernia is caused by lack of closure of pleuroperitoneal canal between 8th- 10th week of fetal life during embryonic development. Etiologically, hernias may be acquired or congenital. Up to 7% of patients suffering from closed thoracoabddominal trauma have a post traumatic diaphragmatic tear, most often on the left side [1]. Upto now, a total of around 100 cases of occult asymptomatic bochdaleks hernia have been reported in literature according to a report published in 2001[2]. According to a medline search, there have been only 32 previous cases in which colon was found to be in thorax [3].

The incidence of late onset Bochdalek hernia has not been clearly determined.Reported rates range from 0.17% to as high as 12.7% [4]. As far as risk factors are concerned, no environmental pathogenic factors have been reported for humans. Congenital diaphragmatic hernia (CDH), alwaysbochdalek hernia has been experimentally induced with thalidomide, vitamin A deficiency, polybromobiphenyls or nitrophen. Bochdalek hernia has been associated with antenatal use of thalidomide, quinine, nitrofenide, antiepileptics & vitamin A deficiency.

Case Report

A 48 year old female, first presented to our hospital with chief complaints of epistaxis along with cervical and lumbar back pain. On general examination the findings were: BMI 22.3kg/m^2, Pulse 82 bpm regular, blood pressure 160/90mmHg, Respiratory rate 12/ min,afebrile, no pallor, edema,icterus or cyanosis.On systemic examination all systems including respiratory system were within normal limits. Patient was managed conservatively for epistaxis. Blood for CBC revealed Hb of 10.8gm/dl and peripheral smear showed 11% blasts. Rest of the CBC, LFT, KFT, FBS, FLP and coagulation profile were within normal limits. Further work up showed 65% plasma cells in bone marrow. Serum protein electrophoresis confirmed the presence of M band and urine was positive for bence jones protein. On basis of these investigations she was diagnosed as a case of multiple

myeloma and was started on tab thalidomide 10 mg OD which was then increased to 100 mg OD. Thyroid function tests revealed that patient was also suffering from hypothyroidism for which she was put on tab eltroxin 75 mcg OD. On subsequent visit and follow up one year later, the patient also received autologous stem cell transplant for multiple myeloma. Two years later the patient again presented with painful lesions over bilateral lower limbs. Lesions were brownish-black purpuric rashes with petechiae around ankle and lower third of leg. She was diagnosed to have pigmented purpuric dermatosis and received tab dapsone 50 mg OD for the same.The patient was on continuous treatment with Tab Thalidomide 100mg OD, Tab Telmisartan 40 mg HS, Tab Eltroxin 75mcg OD, Tab Folvite 2.5 mg OD, when she again presented to our hospital with chief complaints of productive cough for one week, fever for one day. Associated complaints by patient included runny nose, generalized bodyache, and loss of appetite. At time of presentation BP was 110/60 mmhg, pulse rate was 110/min, respiratory rate was 20/ min and temperature was 100.9 degrees Fahrenheit. On examination the patient had mild epigastric tenderness with reduced chest expansion and breath sounds on left side. Routine blood investigations done were within normal limits however X-Ray chest PA view showed stomach with air fluid level to be lifted upwards and seen in Para-vertebral region with left basal lung consolidation, suggestive of diaphragmatic hernia(Figure 1).

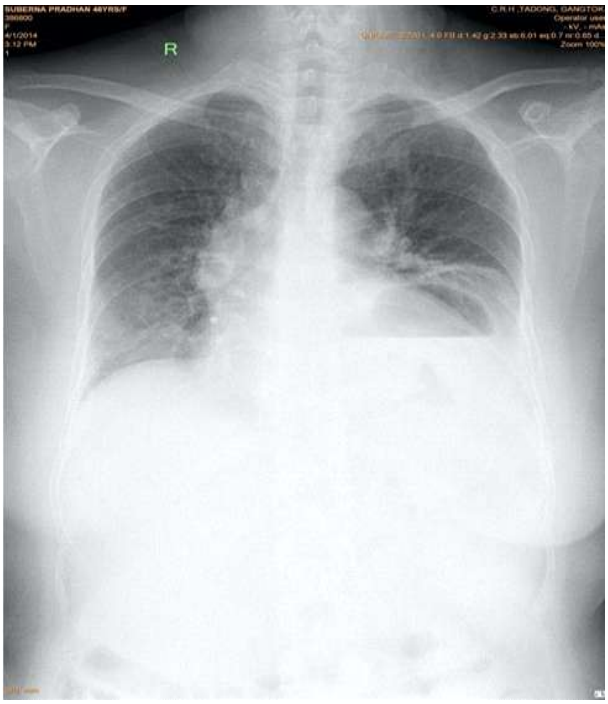

Fig. 1:

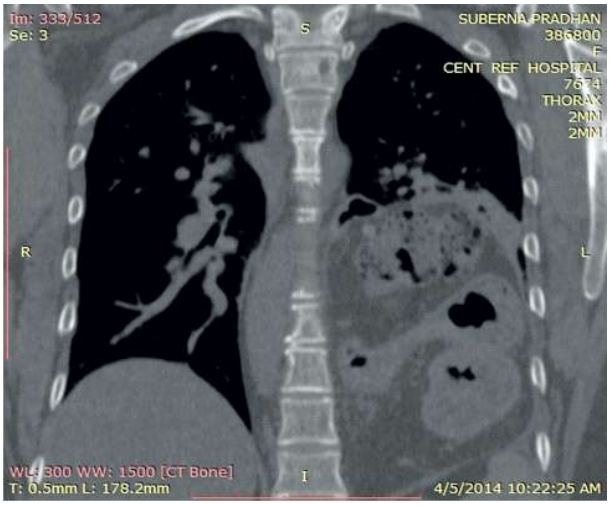

Fig. 2:

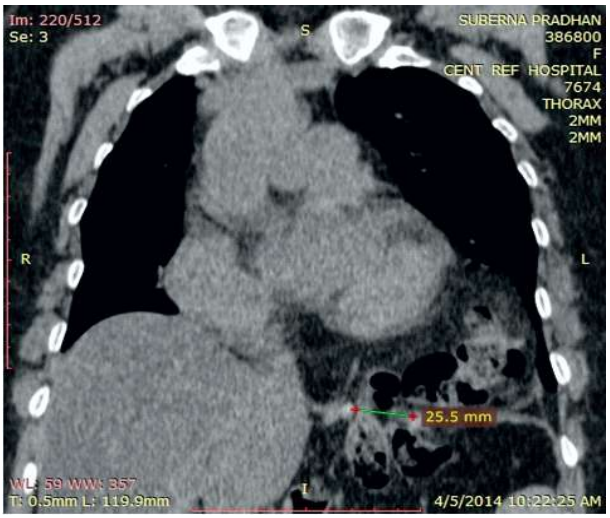

Fig. 3:

CT chest was done which revealed a large size defect in left dome of diaphragm, through which multiple loops of gastro- intestinal tracts were seen to herniate into the chest. The defect in the left dome of diaphragm was placed posteromedially. Herniated contents were notably causing consolidation of left lower lobe (posterior segment) and were displacing the mediastinum along with descending aorta and oesophagus towards right side. Heart was also displaced upwards and towards right side. (Figure 2,3) Above findings confirmed the diagnosis of left diaphragmatic hernia (Bochdalek's Hernia).

Discussion

Bochdalek hernia accounts for approximately 81% of all congenital diaphragmatic hernia. Its

presentation in adult is a rare finding and there are usually two typical clinical presentations in this age group; an incidental finding during X-ray examinations performed for symptoms not related to hernia or when symptoms develop as a result of incarceration, strangulation and visceral rupture inside chest cavity.

Here we present a patient with multiple myeloma, hypertension and hypothyroidism on treatment with tab thalidomide 100mg, tab telmisartan 40mg and tab eltroxin 75mcg, initially with respiratory and gastrointestinal systems within normal limits, complaining of fever, cough and mild epigastric tenderness. There was no history of trauma, lifting of heavy weights, of prolonged respiratory illness or antenatal use of thalidomide by her mother. No history of night blindness was noted. Increased intra-abdominal pressure and thoracic depression may be significant factors for the development of later hernias (in the adult or the elderly). Thus, in obese patients or subjects with kyphoscoliotic deviation, repeated high abdominal pressure events, as in vomiting or coughing may affect reduced-resistance areas in the diaphragm.

The diagnosis of Bochdalek's hernia in adults is not easy, and on couple of occasions has been misdiagnosed as pneumothorax and managed initially by chest tube drainage resulting in feculent discharge from chest and delayed recovery of hernia.The diagnosis of Bochdalek's hernia was confirmed with subsequent radiological examination of X-ray chest and CT scan chest.

Conclusion

Bochdalek's hernia is an uncommon variant of diaphragmatic hernia in adults and symptomatic cases are even rarer. Associations of Bochdalek's hernia with thalidomide as a risk factor has only been seen experimentally, though there have been no case reports till date for proving this theory.After considering all possibilities, we suspectthat the possible cause of this new onset diaphragmatic hernia (Bochdalek's hernia) could be thalidomide intakeas no other risk factors were present in this case.

References

1. L. A. Arráez-Aybar, C. C. González-Gómezand A. J. Torres-García. Morgagni-Larrey parasternal diaphragmatic hernia in the adult ; REV ESP ENFERM DIG (Madrid) 2009; 101(5):357-366.
2. A.Kokacusak, S.Arikan, O.Senturk, A.F.Yucel; bochdalek's hernia in adult with colonic necrosis; hernia 2005; 9:284-287.
3. Ameet Kumar, VikasMaheshwari, TS Rama krishnanand SamareshSahu; Caecal perforation with faecal peritonitis – unusual presentation of Bochdalek hernia in an adult: a case report and review of literature; World Journal of Emergency Surgery 2009; 4:16.
4. Mohammed Zak Rajput, Piero Marco Fisichella; An 81-year-old gentleman with symptomatic Bochdalekhernia; World J Gastrointest Surg 2013 July 27; 5(7):222-223.