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Spontaneous diaphragmatic rupture following neoadjuvant chemotherapy and cytoreductive surgery in malignant pleural mesothelioma: A case report and review of the literature

Margherita Cattaneo*, Paolo Mendogni, Francesco Damarco, Davide Tosi

Fondazione IRCCS Ca' Granda Ospedale Maggiore Policlinico, U.O. Thoracic Surgery and Lung Transplantation, Via Francesco Sforza, 35, 20122, Milan, MI, Italy

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ABSTRACT

INTRODUCTION: Diaphragmatic rupture (DR) is an acquired diaphragmatic defect that can cause herniation of abdominal organs into the chest. It is usually a trauma-related lesion, but rarely it can occur spontaneously. Every DR with abdominal herniation should be considered a surgical emergency.

PRESENTATION OF CASE: A 61-year-old male patient, with previous exposure to asbestos, was diagnosed of Stage Ib malignant pleural mesothelioma (MPM). He underwent neo-adjuvant chemotherapy (three cycle of cisplatin-pemetrexed combination) and a cytoreductive surgery with pleurectomy/decortication. Post-operative course was characterized by prolonged air-leakage (PAL). After three months, during a follow-up CT-scan, a spontaneous diaphragmatic rupture (SDR) with gastric herniation was detected and treated by a laparoscopic diaphragmatic repair and suture.

DISCUSSION: Spontaneous diaphragmatic rupture (SDR) is an extremely rare injury of the diaphragm (less than 1% of all DR). In this case, potential predisposing factors for SDR could be: presence of diaphragmatic "locus minoris resistentiae" due to thinning of the diaphragm and increase tissue fragility after neo-adjuvant chemotherapy and diaphragmatic pleural stripping; increased thoraco-abdominal pressure gradient due to PAL and residual pleural space. Thus, we confirmed the feasibility and safety of the laparoscopic approach.

CONCLUSION: We highlight the multifactor etiopathology, the challenging diagnosis and the importance of a prompt treatment of SDR.

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1. Introduction

Diaphragm is a thin fibromuscular structure that divides the thoracic cavity from the abdomen and one of the major muscles involved in breathing. Its surfaces are covered by three serous membranes: pleura and pericardium on the upper face and peritoneum on the lower one. Due to the negative pressure of the intrapleural space, defects in the diaphragm permit herniation of abdominal organs into the chest. These defects can be congenital or acquired, such as diaphragmatic rupture (DR). Any DR with diaphragmatic herniation could be considered a surgical emergency requiring a multi-disciplinary approach for diagnosis and management.

We report the case of a spontaneous diaphragmatic rupture (SDR) with diaphragmatic herniation following surgical pleurectomy/decortication in a patient affected by malignant pleural mesothelioma (MPM).

This work has been reported in accordance to the Surgical CAse REport (SCARE) guidelines.

2. Presentation of case

A 61-year-old male patient, with previous exposure to asbestos, was referred to our Department by a peripheral hospital with a suspected diagnosis of MPM, after he underwent wedge resection and pleural scarification for a left pneumothorax in June 2019. Radiological assays demonstrated the presence of left PET-positive pleural thickening without pleural effusion nor other PET hypercapitation (Fig. 1). After Multidisciplinary Team revision of the case, in July 2019, the patient underwent left VATS in order to perform adequate pleural biopsies and talc pleurodesis. The histology confirmed MPM, epithelioid subtype, infiltrating the endothoracic fascia, Stage Ib (pT3N0) according to the 8th Edition of TNM [1].

According to the therapeutic protocol of our Centre, the patient received three cycle of neo-adjuvant chemotherapy with cisplatin (CDDP) and pemetrexed (till September 2019) followed by complete radiological re-staging: CT-scan imagines of the

* Corresponding author.

E-mail address: margherita.cattaneo@unimi.it (M. Cattaneo).

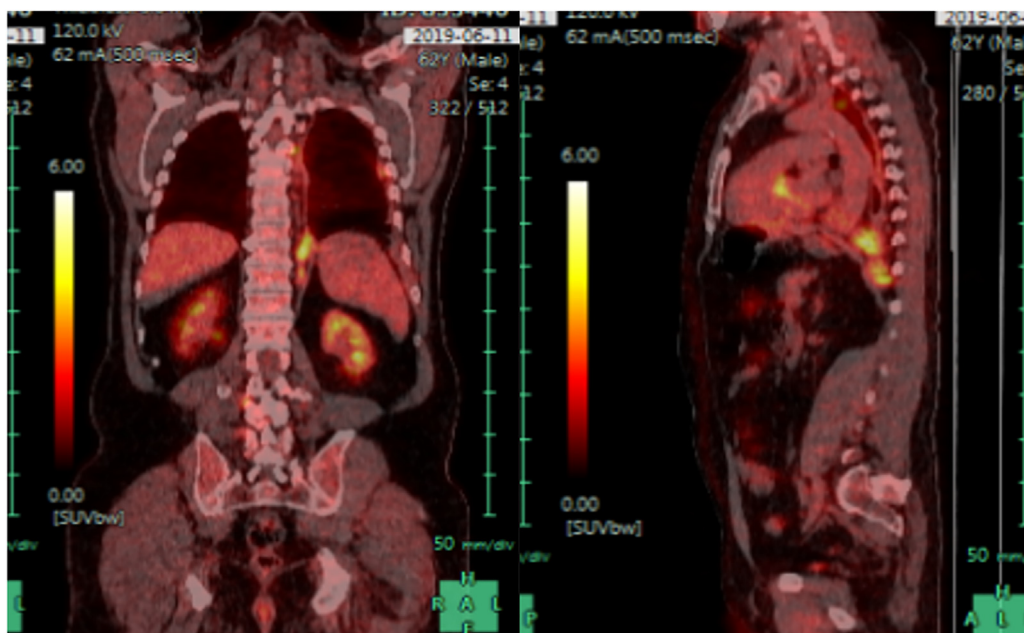


Fig. 1. FDG-PET total body showing pre-treatment status of the malignant pleural mesothelioma.

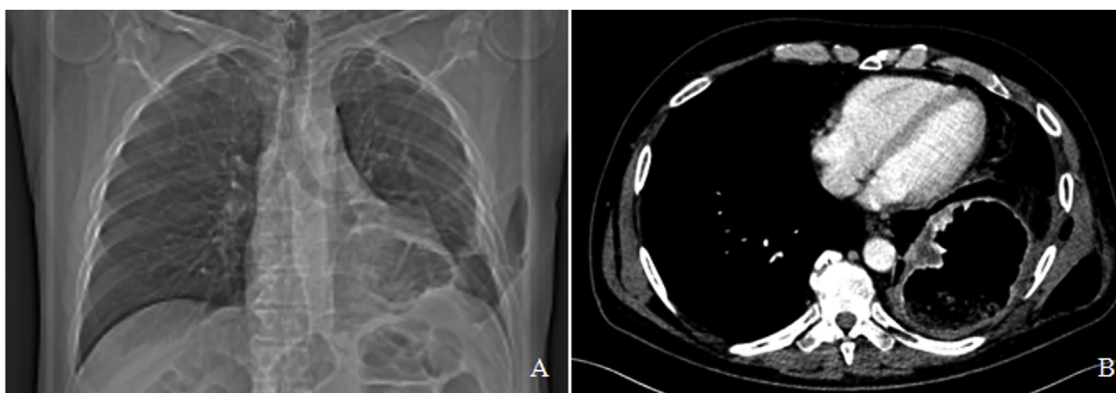


Fig. 2. Thoracic CT-scan showing DR with gastric herniation into the thorax. A: Coronal CT-scan. B: Axial CT-scan.

chest appeared similar to the pre-treatment images, while FDG-PET showed a complete metabolic pleural response. Thus, in November 2019, the patient underwent a cytoreductive surgery with pleurectomy/decortication through a posterolateral thoracotomy; the majority of visceral and costo-vertebral parietal pleura was removed such as diaphragmatic pleura. Multiple pathological plates were localized on the diaphragmatic pleura, as seen during the previous VATS for pleural biopsies, without invasion nor perforation of the diaphragmatic muscular fibers; therefore, a complete left diaphragmatic pleural stripping was performed, with complete preservation of fibromuscular continuity.

Postoperative course was complicated by prolonged air-leakage that resolved spontaneously; chest tube was removed on the 20th post-operative day.

Three months later patient referred new onset of dysphagia and gastroesophageal reflux; thoracic CT-scan identified a 4.5 cm diaphragmatic laceration on the posterior left crus with stomach herniation into the chest, without any sign of incarceration (Fig. 2).

Because of the onset of vomiting and epigastric pain (suggesting a torsion of the gastric hernia) two weeks later, patient acceded to the operating theatre in an emergency setting; a laparoscopic approach was chosen to perform diaphragmatic herniation reduction and injury repair with direct suture. The patient could

start drinking and eating the day after and he was discharged without any complications on post-operative day 4. At the last clinico-radiological follow-up, in June 2020, we confirmed the diaphragmatic integrity and continence and a stable, controlled oncological burden.

3. Discussion

The most common cause of diaphragmatic rupture is trauma, both blunt (in the 75% of the cases) and penetrating (25%) [2]. Spontaneous diaphragmatic rupture (SDR) is an extremely rare injury of the diaphragm (less than 1% of all DR) linked to increased thoracoabdominal pressure gradient without direct trauma, such as heavy physical effort, childbirth, emesis, defecation and coughing [3]. There are two types of SDR described in Literature: type 1, in which the chest wall remains intact; type 2, in which abdominal structures pass through the diaphragm and chest wall [4].

There are no pathognomonic symptoms or signs of SDR and often they are related to the presence of a diaphragmatic herniation, such as compression of the lung, or complications of the hernia itself. Thus, the most common symptoms are dyspnoea, epigastric pain, nausea and vomiting [5].

In our case, we suggest that neo-adjuvant chemotherapy and surgical procedure (with the stripping of the diaphragmatic pleura) could have led to a thinning of the diaphragm, increase tissue fragility and probably to the creation of a “locus minoris resistentiae”. On this stressed setting, then, prolonged air-leakage and the presence of a residual pleural space may have induced a loss of balance between abdominal and thoracic pressures, directly evolving in an SDR.

Early recognition of a DR is crucial to prevent morbidity and mortality; CT-scan is considered the gold standard for its diagnosis, with accuracy ranging from 50% to 78% [6]. DR is always to be considered a surgical emergency which repair is mandatory; complications, such as incarceration, strangulation or perforation, can arise easily, especially in case of delayed diagnosis [7]. Due to the low incidence of this pathology, it is not possible to define the best management, however mini-invasive laparoscopic approach has been reported as safe and feasible [8].

4. Conclusion

This is the first reported case of spontaneous diaphragmatic rupture in a patient affected by malignant pleural mesothelioma treated by chemotherapy and cytoreductive surgery.

Herein we highlight the importance of a multidisciplinary and specialized approach for DR, a rare disease with challenging diagnosis and treatment due to high-risk complications development.

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Ethical approval

Ethical Approval was not necessary for this study. We obtained written patient consent to publication.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author's contribution

Margherita Cattaneo: data collection, literature search, study concept and writing of the article.

Paolo Mendogni: supervision, review and editing of the article.

Francesco Damarco: literature search, conceptualization, and validation of the article.

Davide Tosi: study concept, contribution to the submission process, review and editing of the article.

All authors read and approved the final manuscript.

Registration of research studies

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Declaration of Competing Interest

The authors report no declarations of interest.

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