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CATAMENIAL PNEUMOTHORAX: A RARE CASE REPORT OF A PATIENT FROM THE BRAZILIAN AMAZON PRESENTING PLEURAL EFFUSION

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Abstract: Catamenial pneumothorax is defined as a spontaneous recurrent pneumothorax that occurs in women of reproductive age in a time course related to menstruation. Catamenial pneumothorax is a challenge for the physician to diagnose and treat due to the infrequency of its occurrence and insufficient understanding of the possible risk factors. In this report, we describe a case of a 42-year-old patient who had an atypical presentation with pleural effusion and not pneumothorax. The patient was submitted to an oophorectomy, and in the postoperative period she developed dyspnea on moderate effort and sporadic episodes of orthopnea. The most interesting factor in this case report is the fact that the patient presented with several episodes of pleural effusion and not pneumothorax, the literature reports that spontaneous pneumothorax is the most common clinical manifestation of thoracic endometriosis, occurring in 72-73% of patients, followed by catamenial hemoptysis, catamenial hemothorax and endometriotic thoracic nodules.

INTRODUCTION

Catamenial pneumothorax is defined as a spontaneous recurrent pneumothorax that occurs in women of reproductive age in a time course related to menstruation(1). It has been known since the 1950s and is regarded as a rare entity(2).

Catamenial pneumothorax has a high incidence of relapses. The precise numbers are incompletely known, as few studies have addressed the question of mid- and long-term outcomes(10)

Catamenial pneumothorax is a challenge for the physician to diagnose and treat due to the infrequency of its occurrence and insufficient understanding of the possible risk factors. In this report, we describe a case of a 42-year-old patient who had an atypical

presentation with pleural effusion and not pneumothorax(3)

CASE REPORT

In January 2021, a female patient, 42 years old, initiated dyspnea on moderate effort, after four months the dyspnea became more intense, associated with strong cramps in the lower abdomen during the menstrual period.

She was admitted to a reference center in the capital of Amazonas state presenting pleural effusion in the right hemithorax. Chest computed tomography showed voluminous pleural effusion to the right determining subtotal collapse of the right lung, displacing the hilo-mediastinal structures to the left with massive pleural effusion and moderate ascites. A relief thoracentesis was performed and one and a half liters of pleural fluid were removed.

On physical examination, the patient was in a regular general state, lucid and oriented in time and space, acyanotic, anicteric, afebrile to the touch, pale, hydrated, and dyspneic to room air. There was no palpable lymph node enlargement. On pulmonary auscultation the vesicular murmur was universally diminished in the right hemithorax and present in the left hemithorax, the patient maintained an oxygen saturation of 98%. On cardiac auscultation the heart rhythm was regular, no heart sounds or murmurs were present, the heart rate was 82 beats per minute and the blood pressure was 131 x 89.

During the hospitalization period, the clinical examination revealed the presence of moderate ascites and a mass in the ovaries, and an endovaginal ultrasound was performed, which revealed endometrial thickening, myometrial nodule that may correspond to myoma, and an ovarian mass that may correspond to a teratoma of the right ovary that was not visualized, also performed an abdominal magnetic resonance imaging that showed accentuated pleural effusion on the right, with signs of deep endometriosis in the posterior compartment of the pelvis, with extensive infiltration of the upper rectum/sigmoid and presence of cysts in the right ovary, one of them with a probable endometrial polyp and presence of uterine leiomyomas.

The patient was submitted to an oophorectomy, and in the postoperative period she developed dyspnea on moderate effort and sporadic episodes of orthopnea. The pathological examination of the ovary revealed ovarian tissue fragments, interspersed with endometrial glands and stroma, and recent and organized hemorrhages, delimiting cystic cavities.

Due to several pictures of pleural effusion, patient performed pleural fluid cytology and pleural skin scraping, the fluid was negative for malignant neoplastic cells, showing a hemorrhagic aspect, with clot present, glucose 48; Protein 6g; Albumin 3g; globulin 3g; amylase 40; A/G 1.0; LDH 751. CITOMETRY: 82 cells/mm^3 where 100% are MNNs, presence of Atypical MNNs cells, Rare Gram positive diplococci, absence of BAAR. Anatomopathology of the pleura showed skeletal muscle tissue and scarce fragments of adipose and connective tissue associated with mixed inflammatory infiltrate with a predominance of lymphocytes with crushing artifacts, evidencing stromal endometriosis with ectopia of the endometrial stroma, with foci of recent hemorrhage and in organization with hemosiderin deposits and relational fibrosis, without morphological signs of malignancy. Patient follows well and stable under the care of thoracic surgery, with no further complications to date.

DISCUSSION

The clinic of catamenial pneumothorax is related to endometriosis, occurs by hormonal changes, due to the presence of functioning endometrial tissue in the thorax. Endometrial implants can be found in the lung parenchyma, visceral pleura , parietal pleura and diaphragm(2,4).

There are three theories that explain how endometrial tissue implantation into the thorax occurs, these being cellular metaplasia, lymphatic or hematogenous embolization from the uterus or pelvis, and retrograde menstruation with subsequent transperitoneal - transdiaphragmatic migration of endometrial tissue. However none of these theories could explain the clinical-pathological presentations of catamenial pneumothorax, thus suggesting that the disease has a multifactorial cause(2,4,5).

The pathophysiology of catamenial pneumothorax can be explained in the menstrual period, in which there is an absence of the cervical mucus plug, thus allowing a communication between the peritoneal cavity and the outside through the uterine cavity and fallopian tubes. Air may be forced into the peritoneum by uterine contractions, physical exertion, or sexual intercourse, and thus may reach the pleural space through diaphragmatic defects due to negative intrathoracic pressure(2,4).

The most common symptoms of catamenial pneumothorax are similar to other types of pneumothorax, with cough, chest pain and shortness of breath being the most common symptoms(4,6). Chest pain usually presents as periscapular or neck pain (diaphragmatic pain). Catamenial pneumothorax may be associated with other manifestations of thoracic endometriosis syndrome such as hemothorax(2,7).

At physical examination, the vesicular murmur may be decreased or absent on the side affected by thoracic endometriosis syndrome (TES)(8). Complementary tests such as chest radiography, computed tomography (CT) to visualize the characteristic lesions of thoracic endometriosis syndrome and transvaginal ultrasonography (USG) may be requested to help in the diagnosis, in order to verify the presence of pelvic endometriosis(1).

The management and treatment of catamenial pneumonia should be multidisciplinary and include surgery, if possible by video-assisted technology, and hormonal treatment. Treatment aims to block hormonal support to existing endometrial implants and prevent new implants.Studies on the use of oral contraceptives, progestins, danazol, and gonadotropin-releasing hormone (GnRH) agonists are all available in the literature, but no controlled trials on the efficacy of these drugs in the setting of catamenial pneumothorax are available(6,9).

CONCLUSION

Catamenial Pneumothorax should be considered in all young women presenting with pneumothorax or hemopneumothorax during menstruation. (report the patient's outcome as reported). The most interesting factor in this case report is the fact that the patient presented with several episodes of pleural effusion and not pneumothorax, the literature reports that spontaneous pneumothorax is the most common clinical manifestation of thoracic endometriosis, occurring in 72-73% of patients, followed by catamenial hemoptysis, catamenial hemothorax and endometriotic thoracic nodules(2,6,11,12). This paper serves as data to demonstrate an atypical presentation of catamenial pneumothoraces, and through further studies propose better diagnostic evaluation measures and future therapies.

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