PLEURAL ENDOMETRIOSIS: A CASE STUDY IN MPIGI DISTRICT, UGANDA.

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Abstract

Background:

A 29-year-old female came into our hospital presenting with shortness of breath, the feeling of fluids splashing into the right chest, right-sided chest pains, and mild non-productive cough. The signs and symptoms were always present monthly at the start of her menses.

Case presentation:

It is estimated that 10% of women of reproductive age have endometriosis where the incidence is noted to be as high as 49% in women with chronic pelvic pain. The most common form of endometriosis is abdominopelvic endometriosis, but there are rare cases where there is the presence of endometrial tissue in the thoracic cavity. Pleural endometriosis is a form of thoracic endometriosis in which the endometrial tissue is found on the pleural membrane.

Results:

The chest X-ray findings revealed a right-sided moderate pleural effusion. The cause of this pleural effusion remained idiopathic. However, since her history of presenting complaints was aligned with her menstrual periods, a pleural tissue biopsy was done, which revealed endometrial tissue implanted on the pleural membrane.

Conclusion:

Pleural endometriosis is a rare but existing form of endometriosis with no specific risk factors or signs and symptoms. This explains why many have misdiagnosed it calling it pneumonia. Clinicians are recommended to take a detailed history from patients regardless of how unrelated some complaints may seem. "It seemed unrelated for a pleural effusion with a cough to become a gynecological issue".

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1. Introduction

Normally, the endometrial tissue is only found lining the uterine cavity as the endometrium. However, there are conditions where this tissue is found in other human body parts other than lining the uterine cavity; this condition is termed as endometriosis (Zondervan, Becker & Missmer, 2020), they added that is estimated that 10% of women of reproductive age have endometriosis where the incidence is noted to be as high as 49% in women with chronic pelvic pain.

The most common form of endometriosis is abdominopelvic endometriosis but there are rare cases where there is presence of endometrial tis-

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sue in the thoracic cavity (mostly in the lung parenchyma, air ways and the pleural membrane) which is called thoracic endometriosis. Pleural endometriosis is a form of thoracic endometriosis in which the endometrial tissue is found on the pleural membrane (Chapron, Marcellin, Borghese, Santulli, 2019).

The exact cause of endometriosis of any kind remains a challenge. However there are three accepted postulates of the mechanism of endometriosis; the first is retrograde menstruation which explains only pelvic endometriosis, the second is of serosal cells metaplasia where the remnants of the Muellerian ducts with its coelomic cells retain their potential to differentiate into endometrial tissue - this postulate explains pelvic, peritoneal and limb bud endometriosis, and the third postulate is vascular and lymphatic embolization which explains endometriosis distant sites (Tindall Jeffcoates' V. R. Principles of Gynaecology 5^{th} edition, pg; 361-70) (Dipalos, Mari-G et al., 1989).

Pleural endometriosis is a very rare form of endometriosis. For example, in Uganda, the first case to be published was in 2001 by Byanyima in her article entitled "Menstruation in an unusual place: a case of thoracic endometriosis in Kampala, Uganda" where a 34-year-old female was found having a right posterior pleural mass that was revealed to be and endometrial component. The manifestation of thoracic endometriosis may be catamenial or not, where the catamenial means a clinical manifestation that occurs around the time of menses (24 hours before the onset of menses to 72 hours after the onset of menses) (Bobbio et al., 2019).

Another case report by Wang, Meng, Li and Xu (2021) titled *Endometriosis-Related Pleural Effusion,* found out that catamenial symptoms were presented in 44.8% patients where dyspnea was presented in 74.6% patients followed by right chest/shoulder pain in 50.7% and cough in 26.9%. In another case report, it was reported that majority of the patients had chest pain (90%), dyspnea (31%), haemoptysis (7%), and cough as less common.

Radiological chest studies (X-ray radiography of the chest and Computed tomography of the chest) are among the baseline investigations in pleural endometriosis where majority of patients are found with catamenial pneumothorax (35.8%), lung bullae (1.7%), and pleural thickening or nodularity (8.6%) (Wang, Meng, Li & Xu 2021). For hemothorax, it is found predominantly to involve the right hemithorax - up to 70% of the cases and with 20% occurring bilaterally (Channabasavaiah & Joseph 2010).

While the named radiological investigations create an impression of the condition, biopsy of the suspected tissue and histology are the confirmatory tests for the diagnosis of endometriosis (Byanyima (2001) with classic histopathologic features including the presence of endometrial glands, stroma, and hemosiderin-laden macrophages (Kawaguchi et al., 2018). Different forms of treating pleural endometriosis do exist including surgical such as thoracocentesis, scrapping of this ectopic tissue and drug therapy with hormonal therapy alone is associated with higher recurrence after discontinuation (Vercellini et al., 2002). Therefore, combined therapy may be used where drugs such as anti-gonadotropin agents are frequently prescribed both pre- and post-operatively with recommended duration for an anti-gonadotropin agent at least 6 to 12 months. Some of the anti-gonadotrophic agents include cyclic or continuous oral contraceptives, dienogest, danazol, cyproterone acetate, and GnRH agonist like leuprolide (Fournel et al., 2018).

2. Case

A 29-year-old female came into our hospital presenting with shortness of breath, the feeling of fluids splashing into the right chest, right-sided chest pains, and mild non-productive cough. The signs and symptoms were always present monthly whenever in her menses, sometimes starting shortly before or during the menstrual periods and would disappear soon after completing her periods.

She reported how the intensity of the signs and symptoms, more so the shortness of breath, had

increased that time around. She had even gone to a certain hospital where they had tapped her pleural fluid several times with a working diagnosis of idiopathic pleural effusion.

2.1. ***Past obstetric/gynecological history:***

She reported having never conceived, and her menstrual cycles were regular though painful with a length of about 4 to 6 days.

2.2. ***Family-social history:***

She is not yet married with no clear job but still staying with her parents who try to take care of her medical bills.

2.3. ***On examination:***

She had fast breathing (respiratory rate 32 breaths per minute) though with relatively normal blood pressure (130/80 mmHg), the pulse was high (110 beats per minute), and there was no pallor of mucous membranes.

Multiple scars in the right triangle of safety, right sub-costal tenderness, dullness on percussion, epigastric tenderness, and loss of breath sound in the right basal lung fields.

2.4. ***Investigations:***

Among other investigations, the attending doctor recommended a Chest X-ray.

2.5. ***Radiological examination - Chest X-ray (Postero anterior projection);***

2.5.1. ***Preparations:***

The client was received in the radiology department, her request form was read and she was prepared for the examination. To prepare the patient:

- She was informed about the examination to allay her anxiety
- She was then given a hospital gown and all radio-opaque objects in the chest area were removed.
- Thereafter, she was positioned well upright, with legs apart, standing facing the vertical bucky where her mid sagittal corresponded with the midsagittal of the image receptor.
- Her arms were positioned to retract the scapula away from the lung fields and with centering done at the inferior border of the scapulae. She was then told to inhale and hold them with a high Kilo-volts - technique, and exposure was made.

2.5.2. ***Radiological findings:***

There was opacification of the lower third of the entire right lung with loss of the right costophrenic angles and creating a horizontal linear interface superiorly with the aerated lung field. The preserved lung fields also demonstrated some areas of hazy linear opacities. There was a mediastinal slight shift to the left side where the distal trachea and the cardiac shadow were seen more to the left than their normal position. The left lung fields appeared normal, the left costo-phrenic and cardio-phrenic angles appeared sharp, and the bonny and soft tissue structures of the chest appeared normal.

2.5.3. ***Radiological impression:***

The above radiological description was in keeping with right-sided moderate pleural effusion. The causes of this pleural effusion remained idiopathic. However, because the Complete Blood count Revealed slightly elevated neutrophils, the tentative diagnosis was Pneumonia.

Final diagnosis: On the other hand**,** since her history of presenting complaints was aligned with menstrual periods, another impression of thoracic endometriosis was developed. This led to further investigations which included Computed Tomography (CT) of the chest and Tissue Biopsy.

The histological results confirmed the presence of endometrial stroma and glands along the basal right parietal pleural membrane. This was conclusive of Pleural Endometriosis.

2.5.4. ***Management of this condition:***

Thoracocentesis was done which is the surgical removal of the abnormal fluid collection from the pleural spaces where 750mls of bloody fluid was removed.

The patient was put on drug therapy, that is leuprolide injection 11.25mg which is a

gonadotropin-releasing hormone agonist and then was referred to a gynecologist for further attention.

3. Discussion:

The 29-year-old female had several times complained of shortness of breath, right sided chest pains, and mild non-productive cough shortly before and during her menses, although she reported that the current episode was more severe than the previous ones. This was in line with the report from Wang, Meng, Li and Xu (2021) where they found out that catamenial symptoms were presented in 44.8% patients where dyspnea was presented in 74.6% patients followed by right chest/shoulder pain in 50.7% and cough in 26.9%.

Furthermore, she reported to have been tapped more than once on her right side to remove the accumulated pleural fluids. This was evidenced by various scars observed in the right triangle of safety on examination. In addition, the X-Ray radiological findings revealed a right pleural effusion where about 750mls of bloody fluid were removed. This was in agreement with Channabasavaiah & Joseph (2010) who also reported that hemothorax is found predominantly to involve the right hemithorax in up to 70% of the cases and with 20% occurring bilaterally. The unsorted question remained, "Why is endometriosis common in the right thoracic area than the left?". The radiological findings didn't reveal any pneumothorax which was contrary to the report by Wang, Meng, Li and Xu (2021), who found out that majority of patients were found with catamenial pneumothorax (35.8%) and lung bullae (1.7%).

However much an X-Ray of the chest was done, it couldn't conclude on the cause of this pleural effusion, but because the features had become chronic and aligned with her menstrual periods, this caused the attending medical officer to request for other investigations including lung and pleural biopsy. This was in line with a report from Byanyima (2001) which stated that radiological investigations create an impression of the condition whereas biopsy of the suspected tissue and histology are the confirmatory tests for the diagnosis of endometriosis.

When the biopsy was taken, the histological results confirmed the presence of endometrial stroma and glands along the basal right parietal pleural membrane which was conclusive of Pleural Endometriosis. Kawaguchi et al. (2018) in their report stated that the classic histopathologic features of thoracic endometriosis include the presence of endometrial glands, stroma, and hemosiderin-laden macrophages.

4. Conclusion:

Pleural endometriosis is a rare but existing form of endometriosis where the endometrial tissue is found implanted within or on the pleural membrane. Like other forms of endometriosis, there are no specific risk factors or signs and symptoms for this condition, and that is why many have misdiagnosed it and treated it as pneumonia with no progress.

The presentation of pleural endometriosis with signs and symptoms commonly aligned with the menstrual cycles helps a clinician to develop an impression of this condition. Therefore, different investigations like radiological and histological can help to arrive at this diagnosis.

The management of this pleural endometriosis is a multi-disciplinary approach that includes; clinical, radiological, surgical, and pharmaceutical modalities, and draining of the accumulated pleural fluids, which give relief to the symptoms. However, the use of hormonal therapy and the scrapping of this tissue surgically have delayed the presentation of this condition once more.

5. Recommendations:

Clinicians are recommended to take a detailed history from the patients because it is through this, that, those different diagnoses can be made in spite of how unrelated some complaints may be. "It seemed unrelated for a pleural effusion with a cough to become a gynecological issue".

For any clinical presentations that are aligned with someone's menstrual periods, among your

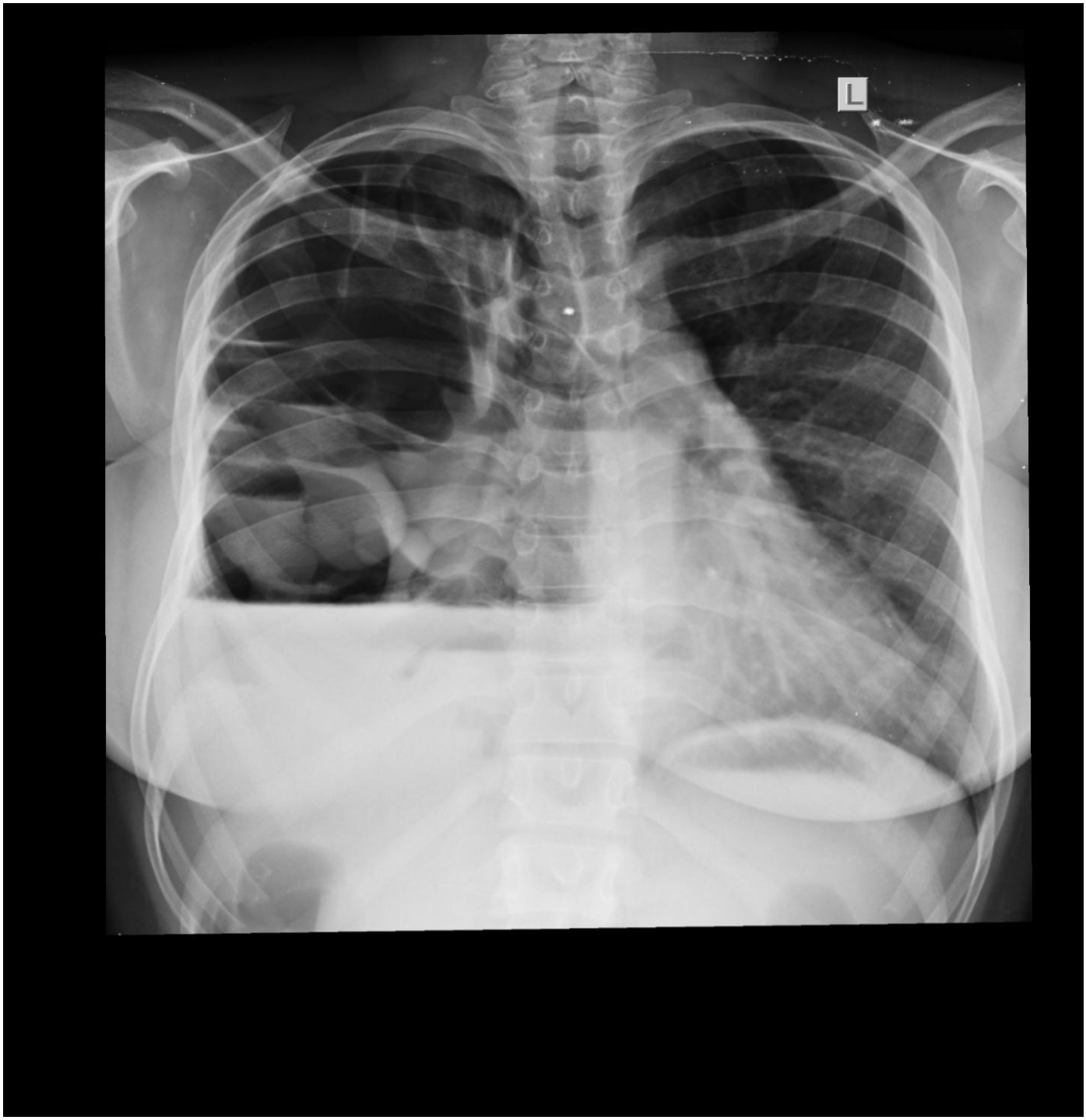

Figure 1: A chest X-ray radiograph of the patient

possibilities, include endometriosis, and with different diagnostic modalities, the final diagnosis can be revealed.

Finally, for any recurring fluid accumulations in the pleural space with no clear diagnosis so far reached, we recommend that a biopsy is done alongside the radiological investigations.

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7. Funding:

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8. Ethics declarations:

With approval from the management of Double Cure Hospital, I then had to seek a consent form from the patient to whom I explained clearly that this is research work that is to help the public gain more knowledge on the case and so there might be no direct gains to her financially. She agreed and consented where the copies of the consent form with the hospital, the patient, and myself.

9. Operational definitions

Endometrium: is the inner lining of the uterine cavity that normally sheds off monthly during menstruation.

Endometriosis: is a condition where there is presence an extra-uterine endometrial tissue.

Pleural: is a double membrane that encases the lung tissue.

Pleural effusion: is the abnormal accumulation of fluids in the pleural cavity.

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