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A female with solitary fibrous tumor pleura mimicking pulmonary sclerosing in low resource setting: A Case Report

--Manuscript Draft--

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Abstract:	Background: Solitary fibrous tumor of pleura (SFTP) is a rare condition. Clinical symptoms and non-specific radiological features in both tumors make preoperative diagnosis difficult to establish. Case presentation: A Javanese 47-year-old female complained of chest pain and shortness of breath which an X-ray and CT-Scan of the thorax showed giant cell metastases in the lung. Signs and symptoms indicate pulmonary sclerosing pneumocytoma (PSP) and the patient underwent thoracic surgery which was first performed with angiographic embolization. Furthermore, an anatomical pathology analysis was performed with suspected SFTP, supported by the IHC test, which found CD34 (+), EMA (-), and S100 (-). Thoracic surgery followed by radiotherapy and chemotherapy is recommended in SFTP patients. Discussion: The SFTP and PSP have non-specific clinical symptoms and radiological features. Anatomic pathology and IHC test are definitive diagnostic tools from SFTP and PSP. Establishing a preoperative diagnosis of SFTP and PSP is quite difficult. Surgical resection is the treatment of choice for both. Conclusion: The comparison of SFTP and PSP can be seen results of the anatomical pathology analysis and IHC test.

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All authors must disclose any financial and personal relationships with other people or organisations that could inappropriately influence (bias) their work. Examples of potential conflicts of interest include employment, consultancies, stock ownership, honoraria, paid expert testimony, patent applications/registrations, and grants or other funding.

Sakina, Isnin Anang Marhana, and Dhihintia Jiwangga Suta Winarno declare that they have no conflict of interest.

Please state any sources of funding for your research

All sources of funding should be declared as an acknowledgement at the end of the text. Authors should declare the role of study sponsors, if any, in the collection, analysis and interpretation of data; in the writing of the manuscript; and in the decision to submit the manuscript for publication. If the study sponsors had no such involvement, the authors should so state.

None.

Ethical Approval

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Author contribution

Please specify the contribution of each author to the paper, e.g. study concept or design, data collection, data analysis or interpretation, writing the paper, others, who have contributed in other ways should be listed as contributors.

Sakina: Data curation, supervision, visualization, investigation, and drafting; **Isnin Anang Marhana:** Conceptualization, methodology, drafting, editing, revising, and reviewing; **Dhihintia Jiwangga Suta Winarno:** Methodology, editing, and revising.

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Isnin Anang Marhana is the person in charge of the publication of our manuscript.

Response to Reviewer

Reviewers' comments:

Reviewer #1: An interesting paper about a common topic in thoracic surgery. Some questions arise from the lecture:

1. You mentioned the FNB of the mass that was unconcluded ... why did you think it could be a PSH? Thd mass was big ... did you perform another biopsy because it could be better to go to surgery knowing the origin of such great mass.

   Author response: we did FOB after FNAB didn't success.

2. You performed pre-operative embolization of the tumour ... is it usual in your experience with all the great thoracic tumours or not? Did you experience any surgical or post-surgical problems?

   Author response: we always had pre-operative embolization of the great thoracic tumors and post-surgical we had no problems.

3. The FSPT could be begnin (most of all) or malign in particular the great masses ... did you analyze the cell mitosis numeber and immunochemistry to clarify the begnin or melignant origin of the tumour? It's important for the recurrence rate of the tumour that was higher in malignant ones

   Author response: It is true that anatomical pathological analysis is very important post-surgical for the prevention of recurrence.

4. Specify the pleural origin of the mass if parietal or visceral it is not clear ...

   Author response: we have add in our manuscript.

Reviewer #2:

1. The authors present an interesting case of a large pleural solitary fibrous tumor (SFTP) which had been radiographically misdiagnosed as a pulmonary sclerosing pneumocytoma. Biopsies had not facilitated a histologic diagnosis, so primary resection was performed after preoperative vascular embolization with curative intent leading to the diagnosis. The case is stated to be the first report of a SFTP in Indonesia.

   Author response: we have revised it in our manuscript.

2. Unfortunately there are some issues concerning case report which make a major revision advisable.

   Author response: we have revised it in our manuscript.

3. Firstly, it is at least since the WHO-Classification of 2015 that pulmonary sclerosing hemangioma should be named pulmonary sclerosing pneumocytoma (PSP), since an origin from pneumocytes II could be confirmed.

   Author response: we have revised it in our manuscript.

4. The abstract is very compact but it would profit from some additional information and more structure to increase readability. IHC tests performed could be mentioned, clinical symptoms are indicated but not specified (Which is o.k. when they are non specific, but then non-specificity should be mentioned), the abbreviation PSH is used but not explained, verbs in past tense and present tense are mixed. The reason for the clinical diagnosis of PSH is not given. It is not made clear, why surgical treatment has priority and what could be alternatives. The conclusion mentions improvement of lung expansion, but that had not been defined as a clinical aim of treatment.

Author response: we have revised abstract in our manuscript.

5. Keyword: Why intrapleural sarcoma? Where is pulmonary sclerosing üneumocytoma?

   Author response: we have revised it in our manuscript.

6. The Highlights are lacking details, for example Highlight 2 could include IHC test : SFTP and PSP can be differentiated using the following IHC test…

   Author response: we have revised it in our manuscript.

7. The Case Presentation gives very limited details about the patient. If she had been in treatment for asthma since childhood, where there older X-Rays or CTs to help judge the timeline of tumor development? Clinical symptoms are stated to be non-characteristic, but they seemed to be relevant, since the patient presented in the emergency department. So what were the symptoms?

   Author response: In a developing country, a patient comes to the hospital when the tumor is in grade III so that it is very difficult for us to follow up.

8. The histopathological results are very limited. Immunohistochemical workup included EMA, S100 (both negative) and CD34 (positive), which seems a little narrow to differentiate other tumors like for example hemangiopericytoma, so more details are needed concerning the diagnosis.

   Author response: we did not perform additional checks due to low resources setting.

9. The Discussion should not only focus on SFTP and PSP but also mention other neoplasms (e.g. hemangiopericytoma, neurogenic tumors) which might be diagnostic

alternatives at the beginning and why they were ruled out. The immunohistochemical workup should be explained in more detail. CD34 might be negative in some SFTP, especially in those in malignant transformation. Moleculargenetic tests have been shown to be useful.

Author response: The results of pathological analysis showed that SFTP was confirmed by the supportive IHC results.

10. The same applies to dynamic CT as a means of differentiation for PSP and SFTP, this should be discussed as it might provide diagnostic help, when biopsies seemingly did not in this case.

Author response: Dynamic CT was not performed due to low resource settiing.

11. In the last few years there have been more publications about higher recurrence rates for SFTP, this should also be discussed, as it is an argument for a longer follow up.

Author response: we have revised it in our manuscript.

12. Alternatives to surgical treatment are not discussed. Preoperative embolization was performed to reduce perioperative risk of bleeding, but complication rates of the procedure are not given.

Author response: we have added it in our manuscript.

13. The Conclusions include information which had not been discussed before (e.g. dynamic CT) or the recommendation of surgical treatment. Preoperative embolization is not mentioned, lung expansion, which is mentioned in the abstracts conclusion, is not discussed.

Author response: we have revised it in our manuscript.

14. References are o.k., but could also be expanded.

Author response: we have revised it in our manuscript.

15. The pictures of the arteriography need more explanation.

Author response: we have added an explanation of the angiography results in figure 3.

16. The SCARE-protocol could be used to focus on the challenges of the case and the patients perspective.

Author response: we have revised it in our manuscript.

17. To conclude, there is a good potential for improvement of the case report, but it should not be published in the present state.

Author response: we have revised it in our manuscript.

Assistant Managing Editor

Please can you make the following changes/checks:

1. Please ensure your case report is compliant with the SCARE Guidelines 2020: http://www.scareguideline.com and submit a completed SCARE 2020 checklist. Please pay particular attention to the following criteria which are often missed:
   - Who performed the procedure? (item 9d)
   - patient perspective (item 12)
   - Drug history, family history including any relevant genetic information, and psychosocial history (item 5d)

- Where relevant - intervention adherence and tolerability (item 10c)
- Post-intervention considerations (item 9f)

Author response: we have added item 9d, item 12, item 5d, item 10c, and item 9f in manuscript base on SCARE guideline 2020.

2. Please also ensure you state that the work has been reported in line with the SCARE 2020 criteria:
   Agha RA, Franchi T, Sohrabi C, Mathew G, for the SCARE Group. The SCARE 2020 Guideline: Updating Consensus Surgical CAse REport (SCARE) Guidelines, International Journal of Surgery 2020;84:226-230.
   Author response: we have added in our manuscript.

3. Please cite the SCARE 2020 paper above in your text in the methods section and the add the reference to your references section.
   Author response: we have cited SCARE 2020 guideline in our manuscript.

4. Please ensure you submit a structured abstract with sub-headings as follows:
   Introduction and importance, Case presentation, clinical Discussion, Conclusion
   Author response: we have compiled our case report according to the guidelines.

5. Can you also please ensure you go through the entire manuscript and check the spelling, grammar and syntax and ensure the language is concise. If you need our author support services, you can access them here: https://www.ijspg.com/services/author-support
   Author response: we have used professional and Grammarly translation services.

6. Please be very clear about what this adds to the existing literature and clearly detail learning points.

   Author response: we have taught it on highlights.

7. Please ensure you submit your work with a Research Registry unique identifying number (UIN) if its first in man i.e. the first time a new device or surgical technique is performed: www.researchregistry.com – it can't progress without being registered. Please ensure you also state your registration UIN in your methods section and reference it including a hyperlink to it if registration is appropriate.

   Author response: not applicable.

8. If you haven't already, please include your "highlights" which are 3-5 bullet points summarising the novel aspects and/or learning points (maximum 85 characters, including spaces, per bullet point).

   Author response: we have revised it in our manuscript.

9. The consent statement in the author form is not suitable. We need a statement like this:

   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.

   Please see consent section in instructions to authors for further information.

   Author response: we have added it in our manuscript.

10. Please ensure any images/figures/photos are suitably anonymised with no patient information or means of identifying the patient.

Author response: we have revised it in our manuscript.

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Author response: we have added it in our manuscript.

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- sources of funding
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Author response: we have added everything in our manuscript

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Author response: we have completed it and attached it with our manuscript.

Highlights

1. Solitary fibrous tumor of pleura (SFTP) and pulmonary sclerosing pneumocytoma (PSP) are difficult to distinguish in a low-resource setting.
2. SFTP and PSP can be differentiated using an immunohistopathology test (cluster of differentiation 34/CD54 was positive).
3. Management of SFTP and PSP are both similar to surgery (thoracotomy with a subcostal incision).

A woman with solitary fibrous tumor pleura mimicking pulmonary sclerosing pneumocytoma in low resource setting: A Case Report

Running head: solitary fibrous tumor pleura

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A female with solitary fibrous tumor pleura mimicking pulmonary sclerosing pneumocytoma in low resource setting: A case report

Running head: solitary fibrous tumor pleura

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A woman with solitary fibrous tumor pleura mimicking pulmonary sclerosing ~~hemangioma~~pneumocytoma in low resource setting: A case report

Abstract

Background: Solitary fibrous tumor of pleura (SFTP) is a rare condition. Clinical symptoms and non-specific radiological features in both tumors make preoperative diagnosis difficult to establish. **Case presentation:** A Javanese 47-year-old ~~woman~~ female complained of chest pain and shortness of breath which an X-ray and CT-Scan of the thorax showed giant cell metastases in the lung. Signs and symptoms indicate pulmonary sclerosing pneumocytoma (PSP) and the patient underwent thoracic surgery which was first performed with angiographic embolization. Furthermore, an anatomical pathology analysis was performed with suspected SFTP, supported by the IHC test, which found CD34 (+), EMA (-), and S100 (-). Thoracic surgery followed by radiotherapy and chemotherapy is recommended in SFTP patients. ~~came to the hospital with suspected PSH with abnormalities on the chest radiograph without clinical complaints of respiratory disorders. The results of tumor resection showed that the tumors were attached to the posterior pleural wall with a size of 12×8×5 cm, which does not resemble PSH image. Immunohistochemical (IHC) tests were performed and the results were SFTP.~~ **Discussion:** The SFTP and ~~pulmonary sclerosing hemangioma (~~PSP~~H)~~ have non-specific clinical symptoms and radiological features. Anatomic pathology and IHC test~~examination~~ are definitive diagnostic tools from SFTP and PSP~~H~~. Establishing a preoperative diagnosis of SFTP and PSP~~H~~ is quite difficult. Surgical resection is the treatment of choice for both. ~~It can be performed either as a therapy or a diagnosis especially in difficult cases.~~ **Conclusion:** The comparison of SFTP and PSP can be seen results of the anatomical pathology analysis and IHC test.~~Clinical improvement of SFTP can be achieved by surgery in which lung expansion can be increased.~~

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Keywords: ~~intrapleural sarcoma,~~ pulmonary sclerosing pneumocytoma, solitary fibrous tumors of pleura, surgery

Introduction

Primary pleural tumors are benign or malignant lesions arising from either the parietal or visceral pleura. The most common of non-mesothelioma primary pleural tumors are solitary fibrous tumors of pleura (SFTP) [1]. In North America, it is estimated that tumor incidence is about 2.8 cases per 100,000 hospitalizations. Malignant mesothelioma is more than 90% of primary pleural tumors. Of the remaining 10%, about 5% are classified as SFTP and the other 5% are various less common primary pleural tumors [2]. Moreover, pleural tumors cause 0.3% to 3.5% thoracic tumors globally [1, 3]. The preoperative differential diagnosis that occurs in patients with SFTP is any mass lesion in the chest, ranging from pulmonary carcinoma to various intrapleural sarcomas [4]. In low resource setting, misdiagnosis is possible between SFTP and pulmonary sclerosing pneumocytoma (PSP) [5] because of the similarity of signs and symptoms and limited investigations [6, 7]. Based on the description above, we are interested in reporting a case of solitary fibrous tumors of pleura using the surgical case report (SCARE) 2020 guideline [8].

Case presentation

A Javanese 47-years-old ~~woman~~ female~~was~~ complained of chest pain and shortness of breath since 3 months ago. Patients experienced significant weight loss without any decrease in appetite. The patient was suspected of having a lung tumor based on physical and radiological examination (Figure 1A) when the patient went to the emergency department for asthma exacerbations. Patients were~~as~~ diagnosed with asthma since childhood. Thoracic CT

scan with contrast appeared to be a solid lesion (39 HU) which by contrast was increasingly enhanced in the next phase (133 HU). The tumor size of ±10.7×14.1×9.72 cm in the upper left hemorrhage was attached to the pleura (Figure 1B). ~~The CT Guiding fine needle aspiration biopsy (FNAB) was examined with non-representation results of the preparation. Anatomical pathology examination of core biopsy results showed no visible signs of malignancy.~~ The examination of fiber optic bronchoscopy (FOB) showed a blunt primary carina, narrowing the distal lumen of the main bronchial due to pressure from the posterior wall, narrowing the left main bronchial lumen caused by the pressure of extra lumen mass. The FOB obtained material for anatomic pathology examination and the results showed no malignant cells (Figure 2).

Embolization was performed, showing the hypervascular mass in the left hemithorax which fed on the left intercostal artery level Th 5-6, the recanalized left bronchial artery, and fine branches of other intercostal artery levels. Post vascularization embolization from the feeding arteries of the superior branches of the left intercostal artery was completely closed. Feeding of the recanalized left bronchial artery and fine branches of other intercostal arteries could not be embolized (Figure 3).

The surgical technique use~~sing~~ an anterolateral thoracotomy approach and enter~~sing~~ through the 5th intercostal space. ~~Tumor resection surgery was performed.~~ The tumor was seen as a round, well-defined tumor, mounted on the posterior thoracic wall with a size of 12×8×5 cm (Figure 4) which anatomical pathology analysis showed tissue sections arranged solidly in irregular patterns, consisting of proliferating oval-spindle-nucleated cells and fine chromatin (SFTP). ~~.~~ The results of the immunohistochemical (IHC) ~~examination~~ test are epithelial membrane antigen (EMA) negative in the membrane and cytoplasm of tumor cells, S100 negative in the cytoplasm of tumor cells, and the cluster of differentiation 34 (CD34) positive in tumor cell membranes. ~~From these results,~~ It was concluded that the tumor was a

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~~solitary fibrous tumor~~SFTP. A radiological examination was performed to evaluate the patient, showing no mass found in the right/left lung/mediastinum (Figure 5). ~~The chest x-ray post-surgery shows chronic inflammation.~~ After surgery, the patient did not feel any complaints. The patient underwent radiotherapy and chemotherapy. One-year postoperative evaluation, the patient had no respiratory complaints and no clinical or radiological signs of recurrence were found.

Discussion

A solitary fibrous pleural tumor is a primary tumor arising from the pleura that can be a benign tumor and has various levels to potentially become malignant [1]. There are two types of primary pleural tumors, namely diffuse and localized. Localized tumors are rare, known as SFTP, which arise from the sub-mesothelial mesenchymal layer. An SFTP occurs in a wide age range (5 to 87 years), often found to occur at the age of the sixth and seventh decades, with quite the same frequency in both sexes [4]. The diagnosis of SFTP and PSP can be distinguished, one of them by using dynamic CT-Scan which dynamic CT-Scan can provide a more accurate picture of the lesion in PSP cases [9]. Dynamic CT-Scan was not performed in our case due to resource constraints. Based on X-ray and CT-scan, giant malignancy is not specific. Support argument diagnosis of SFTP base FOB with malignancy results. The IHC test in SFTP included a primary positive CD34 result, followed by CD99, cytokeratin (S100), and epithelial membrane antigen (EMA) [10]. CD34 is the most frequently used biomarker for SFTP, which in PSP patients the IHC examination was seen for TTF-1, EMA, and pan-cytokeratin immunostaining

~~A chest radiograph is a simple initial diagnostic test for SFTP, which is not specific but can show the presence of a mass in the chest. The SFTP has variously sized and generally has a clear border. The mass is usually located near the periphery of the lung. Tumors that arise as~~

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~~a parietal chest wall mass usually have at least one obtuse angle with the pleural surface, which is a computed tomographic scan image. The picture shows a benign SFTP of 3.3 cm in diameter attached to the parietal pleura on the third right costal which pleural mass with the pleural surface. Unfortunately, only one third of the parietal pleura based mass shows obtuse angles [4].~~

~~In that case, CT shows the tumor attached to the pleura. Extra pleural mass should be seen in the PSH, but this does not rule out the possibility of a PSH. Solitary tumor features are also often found in PSH. In the picture, X ray images are generally delimited smooth, and generally do not stick to the pleura. Heterogeneous features, in contrast, support the diagnosis of hemangiomas in this case. In most cases, an immunohistochemical examination is needed to establish a diagnosis of SFTP and rule out differential diagnoses [7 9]. In addition, a dynamic CT is more accurate to determine the diagnosis of PSH or SFTP [2]. Immunohistochemistry can also be very useful in distinguishing SFTP from mesothelioma and sarcoma. The CD34 is the most commonly used IHC. In PSH, these cells react with TTF 1, EMA, and immunostaining pan cytokeratin. Tumor cells include typical superficial layer cells and underlying round cells, which may originate from pulmonary epithelial cells that are characterized by positive staining of epithelial signs, such as EMA and TTF 1~~ [11-13].

~~The preoperative differential diagnosis in patients with SFTP is any mass in the chest. Sclerosis hemangioma is often considered a solid tumor even though it is not specifically stated the type of tumor. In this patient, the results of the first CT scan concluded that the pulmonary mass may be hemangioma or pleural fibroma [4, 12]. Surgical procedures can be performed safely without a preoperative diagnosis [9]. In almost all cases, the main treatment of SFTP is surgical resection [8]. The use of preoperative angiographic embolization is now widely accepted to reduce the occurrence of intraoperative bleeding. Embolization is usually performed 24 72 hours before resection to help hemostasis during surgery [13].~~In cases of

SFTP, surgery is the preferred action which in cases of malignancy can be surgical procedures performed safely without a preoperative diagnosis [11]. The surgical procedure chosen was thoracotomy with an intercostal incision which is a surgical technique that is effectively removing the giant tumor in the lung [14]. Before surgery, angiographic embolization is now widely accepted to reduce the occurrence of intraoperative bleeding. Embolization is usually performed 24-72 hours before resection to help hemostasis during surgery [15]. Post-surgery, patients are at risk for pain and atelectasis so they need to get special attention for pain management and physiotherapy [16]. Meanwhile, some literature recommends that patients undergo radiotherapy or chemotherapy post-surgery [17, 18].

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Conclusion

A Javanese 47-years-old female complained of chest pain and shortness of breath which an X-ray and CT-Scan of the thorax showed giant cell metastases in the lung. Signs and symptoms indicate PSP and the patient underwent thoracic surgery which was first performed with angiographic embolization. Furthermore, an anatomical pathology analysis was performed with suspected SFTP, supported by the IHC test, which found CD34 (+), EMA (-), and S100 (-). Thoracic surgery followed by radiotherapy and chemotherapy is recommended in SFTP patients. The comparison of SFTP and PSP can be seen results of the anatomical pathology analysis and IHC test.~~SFTP and PSH cannot be distinguished based on history and physical examination. The radiological examination that can distinguish the two is a dynamic CT examination. Surgical resection can be performed on both of them to obtain histopathological specimens and as a therapy. In most SFTP and PSH cases, the diagnosis can be determined by anatomic pathology examination of tissue obtained from tumor resection surgery. SFTP and PSH can be cured by total resection surgery. Therefore, surgery can be carried out even without a definitive preoperative diagnosis.~~

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.

Guarantor

Isnin Anang Marhana is the person in charge of the publication of our manuscript.

Provenance and peer review

Not commissioned, externally peer-reviewed

Conflict of interest

Sakina, Isnin Anang Marhana, and Dhihintia Jiwangga Suta Winarno declare that they have no conflict of interest.

Sources of funding

None.

Ethical approval

Not applicable.

Author contribution

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Sakina: Data curation, supervision, visualization, investigation, and drafting; **Isnin Anang Marhana:** Conceptualization, methodology, drafting, editing, revising, and reviewing; **Dhihintia Jiwangga Suta Winarno:** Methodology, editing, and revising.

Research registration

Not applicable.

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References

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Figure 1. A) X-ray and B) preoperative CT-Scan of the lung.

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Figure 4. Size tumor 12×8×5 cm.

Figure 5. Postoperative radiological examination.

A female~~woman~~ with solitary fibrous tumor pleura mimicking pulmonary sclerosing ~~hemangioma~~pneumocytoma in low resource setting: A case report

Abstract

Background: Solitary fibrous tumor of pleura (SFTP) is a rare condition. Clinical symptoms and non-specific radiological features in both tumors make preoperative diagnosis difficult to establish. **Case presentation:** A Javanese 47-year-old ~~woman~~ female complained of chest pain and shortness of breath which an X-ray and CT-Scan of the thorax showed giant cell metastases in the lung. Signs and symptoms indicate pulmonary sclerosing pneumocytoma (PSP) and the patient underwent thoracic surgery which was first performed with angiographic embolization. Furthermore, an anatomical pathology analysis was performed with suspected SFTP, supported by the IHC test, which found CD34 (+), EMA (-), and S100 (-). Thoracic surgery followed by radiotherapy and chemotherapy is recommended in SFTP patients. ~~came to the hospital with suspected PSH with abnormalities on the chest radiograph without clinical complaints of respiratory disorders. The results of tumor resection showed that the tumors were attached to the posterior pleural wall with a size of 12×8×5 cm, which does not resemble PSH image. Immunohistochemical (IHC) tests were performed and the results were SFTP.~~ **Discussion:** The SFTP and ~~pulmonary sclerosing hemangioma (~~PSP~~H)~~ have non-specific clinical symptoms and radiological features. Anatomic pathology and IHC test~~examination~~ are definitive diagnostic tools from SFTP and PSP~~H~~. Establishing a preoperative diagnosis of SFTP and PSP~~H~~ is quite difficult. Surgical resection is the treatment of choice for both. ~~It can be performed either as a therapy or a diagnosis especially in difficult cases.~~ **Conclusion:** The comparison of SFTP and PSP can be seen results of the anatomical pathology analysis and IHC test.~~Clinical improvement of SFTP can be achieved by surgery in which lung expansion can be increased.~~

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Keywords: ~~intrapleural sarcoma,~~ pulmonary sclerosing pneumocytoma, solitary fibrous tumors of pleura, thoracotomy with intercostal incision~~surgery~~

Introduction

Primary pleural tumors are benign or malignant lesions arising from either the parietal or visceral pleura. The most common of non-mesothelioma primary pleural tumors are solitary fibrous tumors of pleura (SFTP) [1]. In North America, it is estimated that tumor incidence is about 2.8 cases per 100,000 hospitalizations. Malignant mesothelioma is more than 90% of primary pleural tumors. Of the remaining 10%, about 5% are classified as SFTP and the other 5% are various less common primary pleural tumors [2]. Moreover, pleural tumors cause 0.3% to 3.5% thoracic tumors globally [1, 3]. The preoperative differential diagnosis that occurs in patients with SFTP is any mass lesion in the chest, ranging from pulmonary carcinoma to various intrapleural sarcomas [4]. In low resource setting, misdiagnosis is possible between SFTP and pulmonary sclerosing pneumocytoma (PSP) [5] because of the similarity of signs and symptoms and limited investigations [6, 7]. Based on the description above, we are interested in reporting a case of solitary fibrous tumors of pleura using the surgical case report (SCARE) 2020 guideline [8].

Case presentation

A Javanese 47-years-old ~~woman~~ female~~was~~ complained of chest pain and shortness of breath since 3 months ago. Patients experienced significant weight loss without any decrease in appetite. The patient was suspected of having a lung tumor based on physical and radiological examination (Figure 1A) when the patient went to the emergency department for asthma exacerbations. Patients were~~as~~ diagnosed with asthma since childhood. Thoracic CT

scan with contrast appeared to be a solid lesion (39 HU) which by contrast was increasingly enhanced in the next phase (133 HU). The tumor size of ±10.7×14.1×9.72 cm in the upper left hemorrhage was attached to the pleura (Figure 1B). ~~The CT Guiding fine needle aspiration biopsy (FNAB) was examined with non-representation results of the preparation. Anatomical pathology examination of core biopsy results showed no visible signs of malignancy.~~ The examination of fiber optic bronchoscopy (FOB) showed a blunt primary carina, narrowing the distal lumen of the main bronchial due to pressure from the posterior wall, narrowing the left main bronchial lumen caused by the pressure of extra lumen mass. The FOB obtained material for anatomic pathology examination and the results showed no malignant cells (Figure 2).

Embolization was performed, showing the hypervascular mass in the left hemithorax which fed on the left intercostal artery level Th 5-6, the recanalized left bronchial artery, and fine branches of other intercostal artery levels. Post vascularization embolization from the feeding arteries of the superior branches of the left intercostal artery was completely closed. Feeding of the recanalized left bronchial artery and fine branches of other intercostal arteries could not be embolized (Figure 3).

The surgical technique use~~sing~~ an anterolateral thoracotomy approach and enter~~sing~~ through the 5th intercostal space. ~~Tumor resection surgery was performed.~~ The tumor was seen as a round, well-defined tumor, mounted on the posterior thoracic wall with a size of 12×8×5 cm (Figure 4) which anatomical pathology analysis showed tissue sections arranged solidly in irregular patterns, consisting of proliferating oval-spindle-nucleated cells and fine chromatin (SFTP). ~~.~~ The results of the immunohistochemical (IHC) ~~examination~~ test are epithelial membrane antigen (EMA) negative in the membrane and cytoplasm of tumor cells, S100 negative in the cytoplasm of tumor cells, and the cluster of differentiation 34 (CD34) positive in tumor cell membranes. ~~From these results,~~ It was concluded that the tumor was a

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~~solitary fibrous tumor~~SFTP. A radiological examination was performed to evaluate the patient, showing no mass found in the right/left lung/mediastinum (Figure 5). ~~The chest x-ray post-surgery shows chronic inflammation.~~ After surgery, the patient did not feel any complaints. The patient underwent radiotherapy and chemotherapy. One-year postoperative evaluation, the patient had no respiratory complaints and no clinical or radiological signs of recurrence were found.

Discussion

A solitary fibrous pleural tumor is a primary tumor arising from the pleura that can be a benign tumor and has various levels to potentially become malignant [1]. There are two types of primary pleural tumors, namely diffuse and localized. Localized tumors are rare, known as SFTP, which arise from the sub-mesothelial mesenchymal layer. An SFTP occurs in a wide age range (5 to 87 years), often found to occur at the age of the sixth and seventh decades, with quite the same frequency in both sexes [4]. The diagnosis of SFTP and PSP can be distinguished, one of them by using dynamic CT-Scan which dynamic CT-Scan can provide a more accurate picture of the lesion in PSP cases [9]. Dynamic CT-Scan was not performed in our case due to resource constraints. Based on X-ray and CT-scan, giant malignancy is not specific. Support argument diagnosis of SFTP base FOB with malignancy results. The IHC test in SFTP included a primary positive CD34 result, followed by CD99, cytokeratin (S100), and epithelial membrane antigen (EMA) [10]. CD34 is the most frequently used biomarker for SFTP, which in PSP patients the IHC examination was seen for TTF-1, EMA, and pan-cytokeratin immunostaining

~~A chest radiograph is a simple initial diagnostic test for SFTP, which is not specific but can show the presence of a mass in the chest. The SFTP has variously sized and generally has a clear border. The mass is usually located near the periphery of the lung. Tumors that arise as~~

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~~a parietal chest wall mass usually have at least one obtuse angle with the pleural surface, which is a computed tomographic scan image. The picture shows a benign SFTP of 3.3 cm in diameter attached to the parietal pleura on the third right costal which pleural mass with the pleural surface. Unfortunately, only one third of the parietal pleura based mass shows obtuse angles [4].~~

~~In that case, CT shows the tumor attached to the pleura. Extra pleural mass should be seen in the PSH, but this does not rule out the possibility of a PSH. Solitary tumor features are also often found in PSH. In the picture, X ray images are generally delimited smooth, and generally do not stick to the pleura. Heterogeneous features, in contrast, support the diagnosis of hemangiomas in this case. In most cases, an immunohistochemical examination is needed to establish a diagnosis of SFTP and rule out differential diagnoses [7 9]. In addition, a dynamic CT is more accurate to determine the diagnosis of PSH or SFTP [2]. Immunohistochemistry can also be very useful in distinguishing SFTP from mesothelioma and sarcoma. The CD34 is the most commonly used IHC. In PSH, these cells react with TTF 1, EMA, and immunostaining pan cytokeratin. Tumor cells include typical superficial layer cells and underlying round cells, which may originate from pulmonary epithelial cells that are characterized by positive staining of epithelial signs, such as EMA and TTF 1~~ [11-13].

~~The preoperative differential diagnosis in patients with SFTP is any mass in the chest. Sclerosis hemangioma is often considered a solid tumor even though it is not specifically stated the type of tumor. In this patient, the results of the first CT scan concluded that the pulmonary mass may be hemangioma or pleural fibroma [4, 12]. Surgical procedures can be performed safely without a preoperative diagnosis [9]. In almost all cases, the main treatment of SFTP is surgical resection [8]. The use of preoperative angiographic embolization is now widely accepted to reduce the occurrence of intraoperative bleeding. Embolization is usually performed 24 72 hours before resection to help hemostasis during surgery [13].~~In cases of

SFTP, surgery is the preferred action which in cases of malignancy can be surgical procedures performed safely without a preoperative diagnosis [11]. The surgical procedure chosen was thoracotomy with an intercostal incision which is a surgical technique that is effectively removing the giant tumor in the lung [14]. Before surgery, angiographic embolization is now widely accepted to reduce the occurrence of intraoperative bleeding. Embolization is usually performed 24-72 hours before resection to help hemostasis during surgery [15]. Post-surgery, patients are at risk for pain and atelectasis so they need to get special attention for pain management and physiotherapy [16]. Meanwhile, some literature recommends that patients undergo radiotherapy or chemotherapy post-surgery [17, 18].

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Conclusion

A Javanese 47-years-old female complained of chest pain and shortness of breath which an X-ray and CT-Scan of the thorax showed giant cell metastases in the lung. Signs and symptoms indicate PSP and the patient underwent thoracic surgery which was first performed with angiographic embolization. Furthermore, an anatomical pathology analysis was performed with suspected SFTP, supported by the IHC test, which found CD34 (+), EMA (-), and S100 (-). Thoracic surgery followed by radiotherapy and chemotherapy is recommended in SFTP patients. The comparison of SFTP and PSP can be seen results of the anatomical pathology analysis and IHC test.~~SFTP and PSH cannot be distinguished based on history and physical examination. The radiological examination that can distinguish the two is a dynamic CT examination. Surgical resection can be performed on both of them to obtain histopathological specimens and as a therapy. In most SFTP and PSH cases, the diagnosis can be determined by anatomic pathology examination of tissue obtained from tumor resection surgery. SFTP and PSH can be cured by total resection surgery. Therefore, surgery can be carried out even without a definitive preoperative diagnosis.~~

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.

Guarantor

Isnin Anang Marhana is the person in charge of the publication of our manuscript.

Provenance and peer review

Not commissioned, externally peer-reviewed

Conflict of interest

Sakina, Isnin Anang Marhana, and Dhihintia Jiwangga Suta Winarno declare that they have no conflict of interest.

Sources of funding

None.

Ethical approval

Not applicable.

Author contribution

Sakina: Data curation, supervision, visualization, investigation, and drafting; **Isnin Anang Marhana:** Conceptualization, methodology, drafting, editing, revising, and reviewing; **Dhihintia Jiwangga Suta Winarno:** Methodology, editing, and revising.

Research registration

Not applicable.

Acknowledgment

We would like to thank our editor "Fis Citra Ariyanto".

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A woman with solitary fibrous tumor pleura mimicking pulmonary sclerosing pneumocytoma in low resource setting: A case report

Abstract

Background: Solitary fibrous tumor of pleura (SFTP) is a rare condition. Clinical symptoms and non-specific radiological features in both tumors make preoperative diagnosis difficult to establish. **Case presentation:** A Javanese 47-year-old female complained of chest pain and shortness of breath which an X-ray and CT-Scan of the thorax showed giant cell metastases in the lung. Signs and symptoms indicate pulmonary sclerosing pneumocytoma (PSP) and the patient underwent thoracic surgery which was first performed with angiographic embolization. Furthermore, an anatomical pathology analysis was performed with suspected SFTP, supported by the IHC test, which found CD34 (+), EMA (-), and S100 (-). Thoracic surgery followed by radiotherapy and chemotherapy is recommended in SFTP patients. **Discussion:** The SFTP and PSP have non-specific clinical symptoms and radiological features. Anatomic pathology and IHC test are definitive diagnostic tools from SFTP and PSP. Establishing a preoperative diagnosis of SFTP and PSP is quite difficult. Surgical resection is the treatment of choice for both. **Conclusion:** The comparison of SFTP and PSP can be seen results of the anatomical pathology analysis and IHC test.

Keywords: pulmonary sclerosing pneumocytoma, solitary fibrous tumors of pleura, surgery

Introduction

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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.

Guarantor

Isnin Anang Marhana is the person in charge of the publication of our manuscript.

Provenance and peer review

Not commissioned, externally peer-reviewed

Conflict of interest

Sakina, Isnin Anang Marhana, and Dhihintia Jiwangga Suta Winarno declare that they have no conflict of interest.

Sources of funding

None.

Ethical approval

Not applicable.

Author contribution

Sakina: Data curation, supervision, visualization, investigation, and drafting; **Isnin Anang Marhana:** Conceptualization, methodology, drafting, editing, revising, and reviewing; **Dhihintia Jiwangga Suta Winarno:** Methodology, editing, and revising.

Research registration

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Figure Legend

Figure 1. A) X-ray and B) preoperative CT-Scan of the lung.

Figure 2. A) Right main bronchus and B) left main bronchus.

Figure 3. Arteriography results recanalized the left bronchial artery.

Figure 4. Size tumor 12×8×5 cm.

Figure 5. Postoperative radiological examination.

A female with solitary fibrous tumor pleura mimicking pulmonary sclerosing pneumocytoma in low resource setting: A case report

Abstract

Background: Solitary fibrous tumor of pleura (SFTP) is a rare condition. Clinical symptoms and non-specific radiological features in both tumors make preoperative diagnosis difficult to establish. **Case presentation:** A Javanese 47-year-old female complained of chest pain and shortness of breath which an X-ray and CT-Scan of the thorax showed giant cell metastases in the lung. Signs and symptoms indicate pulmonary sclerosing pneumocytoma (PSP) and the patient underwent thoracic surgery which was first performed with angiographic embolization. Furthermore, an anatomical pathology analysis was performed with suspected SFTP, supported by the IHC test, which found CD34 (+), EMA (-), and S100 (-). Thoracic surgery followed by radiotherapy and chemotherapy is recommended in SFTP patients. **Discussion:** The SFTP and PSP have non-specific clinical symptoms and radiological features. Anatomic pathology and IHC test are definitive diagnostic tools from SFTP and PSP. Establishing a preoperative diagnosis of SFTP and PSP is quite difficult. Surgical resection is the treatment of choice for both. **Conclusion:** The comparison of SFTP and PSP can be seen results of the anatomical pathology analysis and IHC test.

Keywords: pulmonary sclerosing pneumocytoma, solitary fibrous tumors of pleura, thoracotomy with intercostal incision

Introduction

Primary pleural tumors are benign or malignant lesions arising from either the parietal or visceral pleura. The most common of non-mesothelioma primary pleural tumors are solitary

fibrous tumors of pleura (SFTP) [1]. In North America, it is estimated that tumor incidence is about 2.8 cases per 100,000 hospitalizations. Malignant mesothelioma is more than 90% of primary pleural tumors. Of the remaining 10%, about 5% are classified as SFTP and the other 5% are various less common primary pleural tumors [2]. Moreover, pleural tumors cause 0.3% to 3.5% thoracic tumors globally [1, 3]. The preoperative differential diagnosis that occurs in patients with SFTP is any mass lesion in the chest, ranging from pulmonary carcinoma to various intrapleural sarcomas [4]. In low resource setting, misdiagnosis is possible between SFTP and pulmonary sclerosing pneumocytoma (PSP) [5] because of the similarity of signs and symptoms and limited investigations [6, 7]. Based on the description above, we are interested in reporting a case of solitary fibrous tumors of pleura using the surgical case report (SCARE) 2020 guideline [8].

Case presentation

A Javanese 47-years-old female complained of chest pain and shortness of breath since 3 months ago. Patients experienced significant weight loss without any decrease in appetite. The patient was suspected of having a lung tumor based on physical and radiological examination (Figure 1A) when the patient went to the emergency department for asthma exacerbations. Patients were diagnosed with asthma since childhood. Thoracic CT scan with contrast appeared to be a solid lesion (39 HU) which by contrast was increasingly enhanced in the next phase (133 HU). The tumor size of ±10.7×14.1×9.72 cm in the upper left hemorrhage was attached to the pleura (Figure 1B). The examination of fiber optic bronchoscopy (FOB) showed a blunt primary carina, narrowing the distal lumen of the main bronchial due to pressure from the posterior wall, narrowing the left main bronchial lumen caused by the pressure of extra lumen mass. The FOB obtained material for anatomic pathology examination and the results showed no malignant cells (Figure 2).

Embolization was performed, showing the hypervascular mass in the left hemithorax which fed on the left intercostal artery level Th 5-6, the recanalized left bronchial artery, and fine branches of other intercostal artery levels. Post vascularization embolization from the feeding arteries of the superior branches of the left intercostal artery was completely closed. Feeding of the recanalized left bronchial artery and fine branches of other intercostal arteries could not be embolized (Figure 3).

The surgical technique uses an anterolateral thoracotomy approach and enters through the 5th intercostal space. The tumor was seen as a round, well-defined tumor, mounted on the posterior thoracic wall with a size of 12×8×5 cm (Figure 4) which anatomical pathology analysis showed tissue sections arranged solidly in irregular patterns, consisting of proliferating oval-spindle-nucleated cells and fine chromatin (SFTP). The results of the immunohistochemical (IHC) test are epithelial membrane antigen (EMA) negative in the membrane and cytoplasm of tumor cells, S100 negative in the cytoplasm of tumor cells, and the cluster of differentiation 34 (CD34) positive in tumor cell membranes. It was concluded that the tumor was a SFTP. A radiological examination was performed to evaluate the patient, showing no mass found in the right/left lung/mediastinum (Figure 5). After surgery, the patient did not feel any complaints. The patient underwent radiotherapy and chemotherapy. One-year postoperative evaluation, the patient had no respiratory complaints and no clinical or radiological signs of recurrence were found.

Discussion

A solitary fibrous pleural tumor is a primary tumor arising from the pleura that can be a benign tumor and has various levels to potentially become malignant [1]. There are two types of primary pleural tumors, namely diffuse and localized. Localized tumors are rare, known as SFTP, which arise from the sub-mesothelial mesenchymal layer. An SFTP occurs in a wide

age range (5 to 87 years), often found to occur at the age of the sixth and seventh decades, with quite the same frequency in both sexes [4]. The diagnosis of SFTP and PSP can be distinguished, one of them by using dynamic CT-Scan which dynamic CT-Scan can provide a more accurate picture of the lesion in PSP cases [9]. Dynamic CT-Scan was not performed in our case due to resource constraints. Based on X-ray and CT-scan, giant malignancy is not specific. Support argument diagnosis of SFTP base FOB with malignancy results. The IHC test in SFTP included a primary positive CD34 result, followed by CD99, cytokeratin (S100), and epithelial membrane antigen (EMA) [10]. CD34 is the most frequently used biomarker for SFTP, which in PSP patients the IHC examination was seen for TTF-1, EMA, and pan-cytokeratin immunostaining [11-13].

In cases of SFTP, surgery is the preferred action which in cases of malignancy can be surgical procedures performed safely without a preoperative diagnosis [11]. The surgical procedure chosen was thoracotomy with an intercostal incision which is a surgical technique that is effectively removing the giant tumor in the lung [14]. Before surgery, angiographic embolization is now widely accepted to reduce the occurrence of intraoperative bleeding. Embolization is usually performed 24-72 hours before resection to help hemostasis during surgery [15]. Post-surgery, patients are at risk for pain and atelectasis so they need to get special attention for pain management and physiotherapy [16]. Meanwhile, some literature recommends that patients undergo radiotherapy or chemotherapy post-surgery [17, 18].

Conclusion

A Javanese 47-years-old female complained of chest pain and shortness of breath which an X-ray and CT-Scan of the thorax showed giant cell metastases in the lung. Signs and symptoms indicate PSP and the patient underwent thoracic surgery which was first performed with angiographic embolization. Furthermore, an anatomical pathology analysis was

performed with suspected SFTP, supported by the IHC test, which found CD34 (+), EMA (-), and S100 (-). Thoracic surgery followed by radiotherapy and chemotherapy is recommended in SFTP patients. The comparison of SFTP and PSP can be seen results of the anatomical pathology analysis and IHC test.

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.

Guarantor

Isnin Anang Marhana is the person in charge of the publication of our manuscript.

Provenance and peer review

Not commissioned, externally peer-reviewed

Conflict of interest

Sakina, Isnin Anang Marhana, and Dhihintia Jiwangga Suta Winarno declare that they have no conflict of interest.

Sources of funding

None.

Ethical approval

Not applicable.

Author contribution

Sakina: Data curation, supervision, visualization, investigation, and drafting; **Isnin Anang Marhana:** Conceptualization, methodology, drafting, editing, revising, and reviewing; **Dhihintia Jiwangga Suta Winarno:** Methodology, editing, and revising.

Research registration

Not applicable.

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Figure 1A

Click here to access/download;Figure;06. Figure 1A.jpg

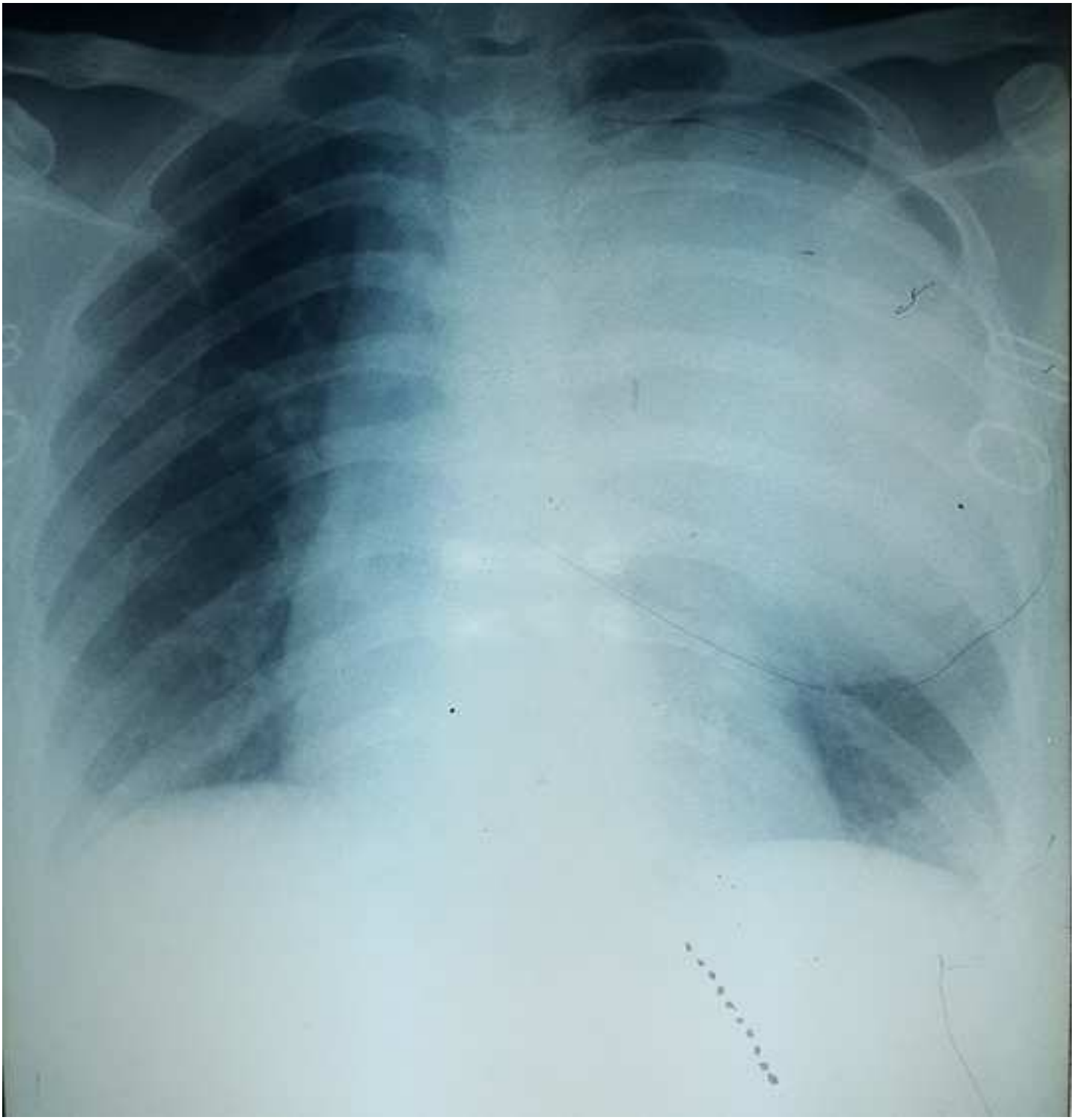

Figure 1B

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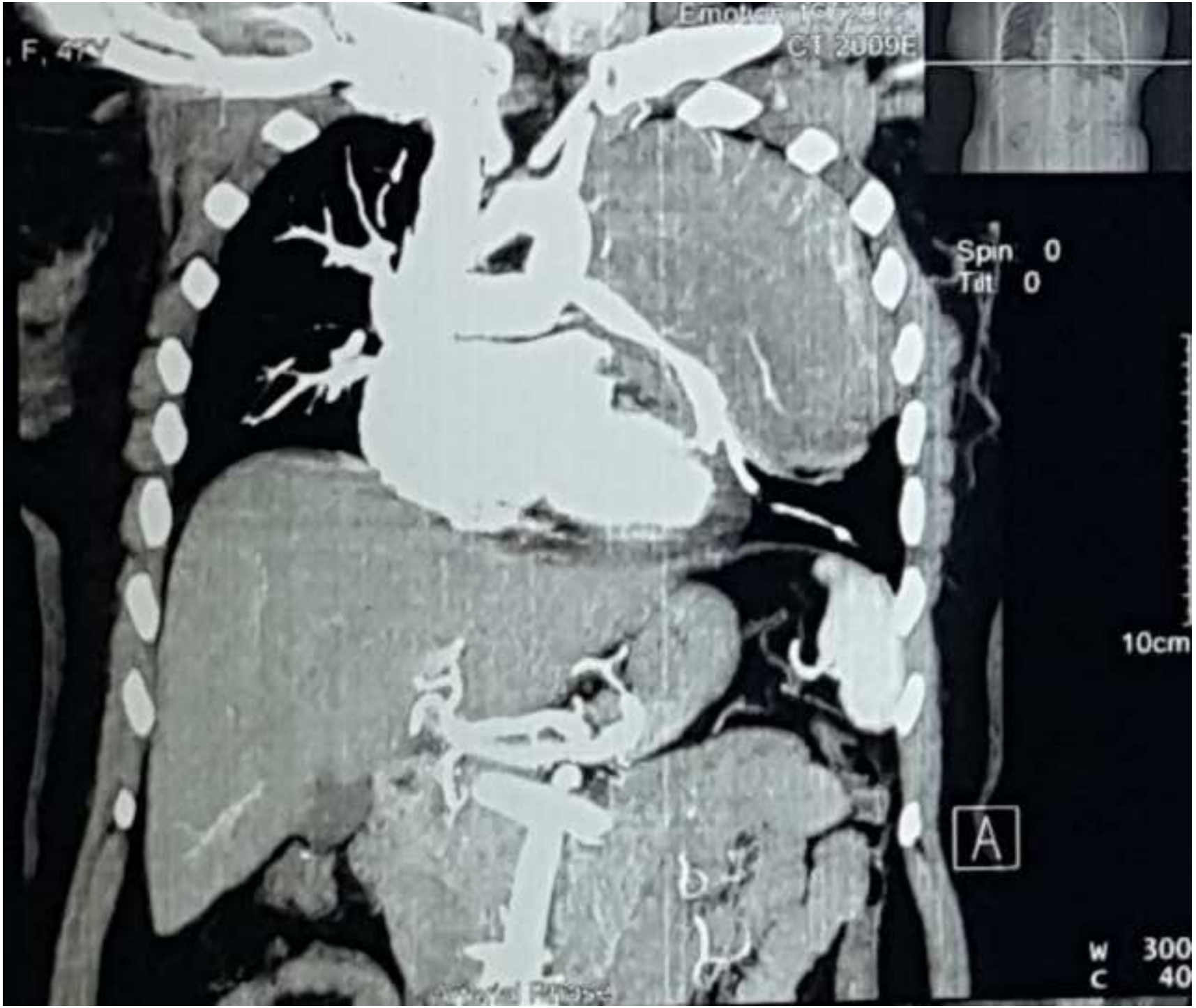

Figure 2

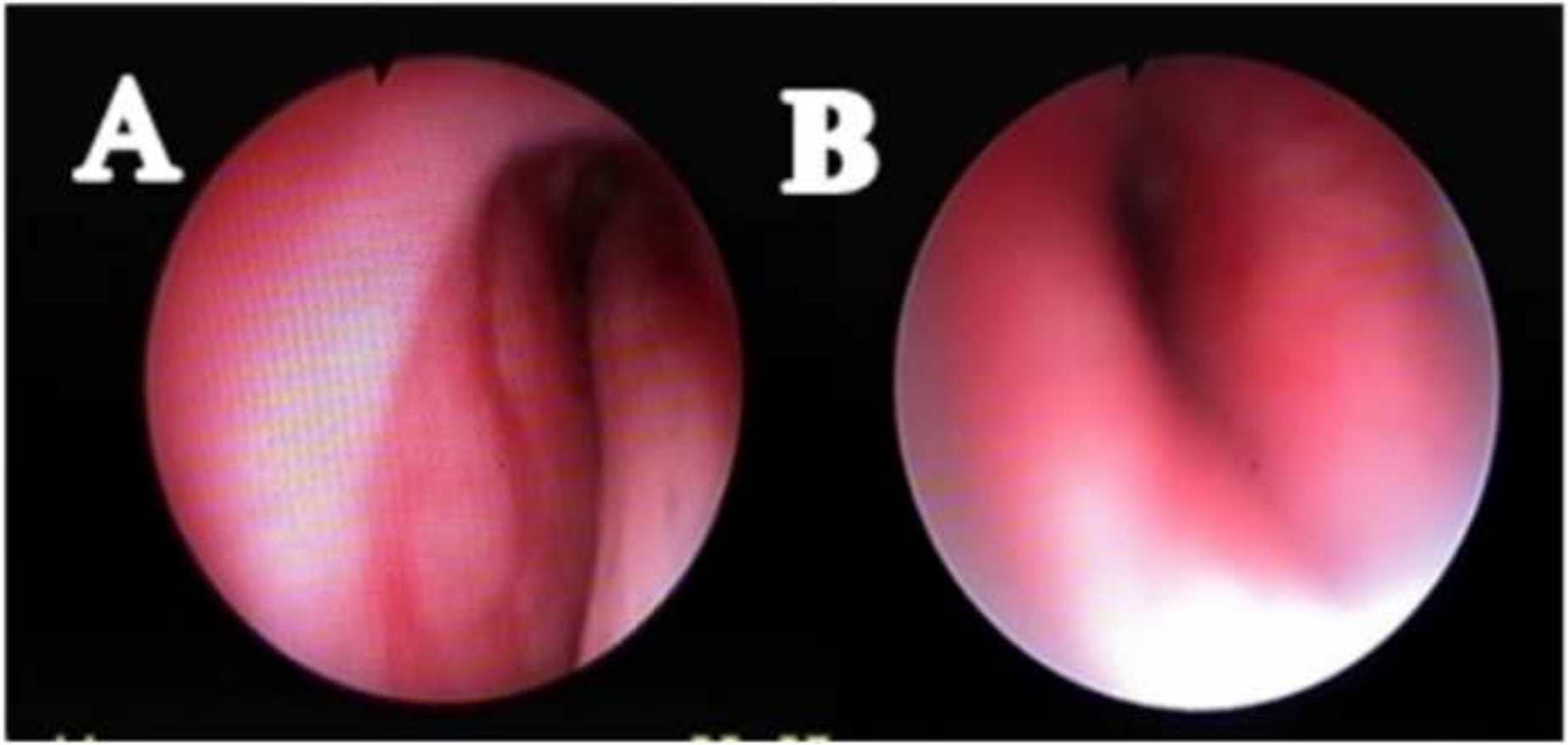

Figure 3A

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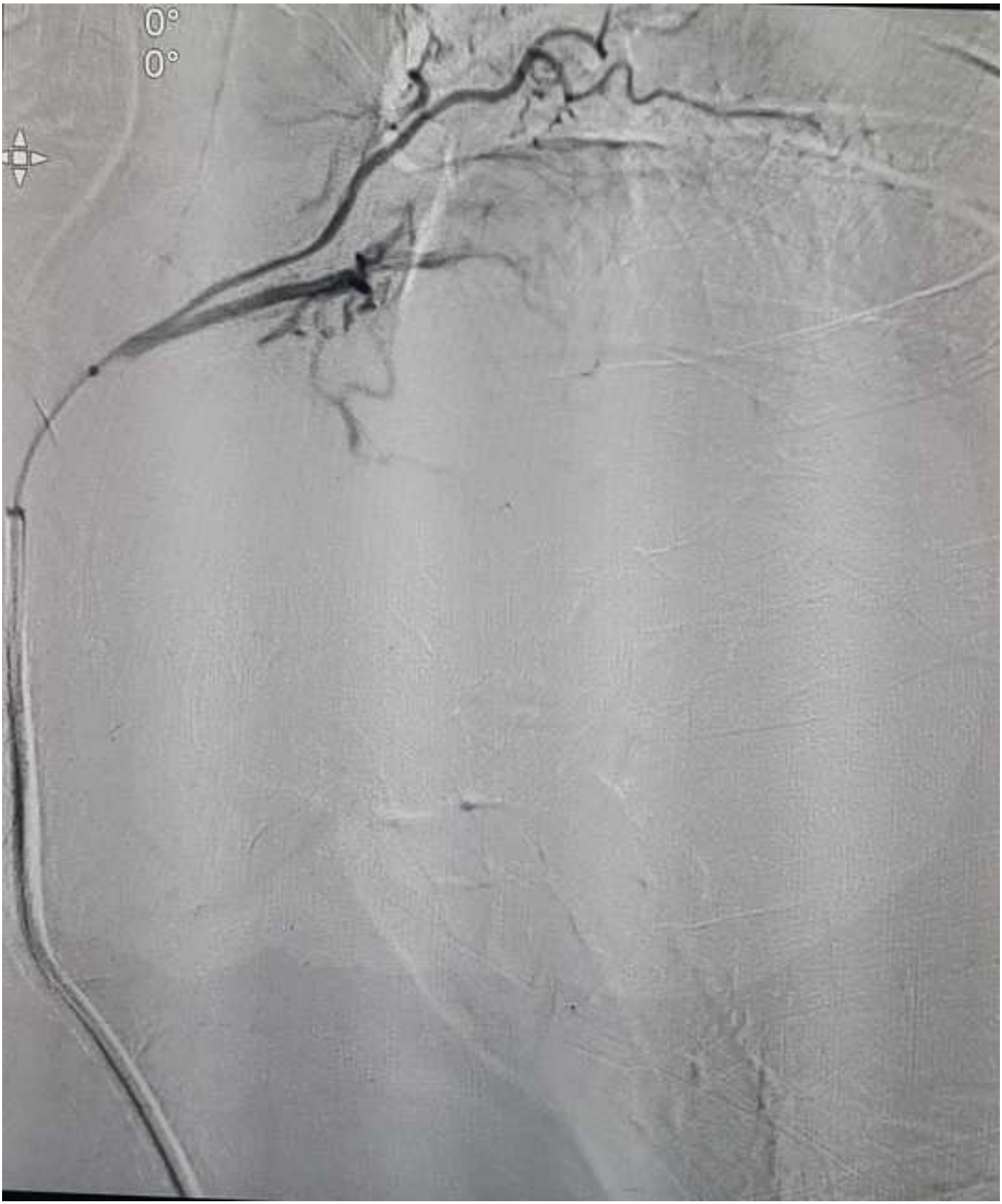

Figure 3B

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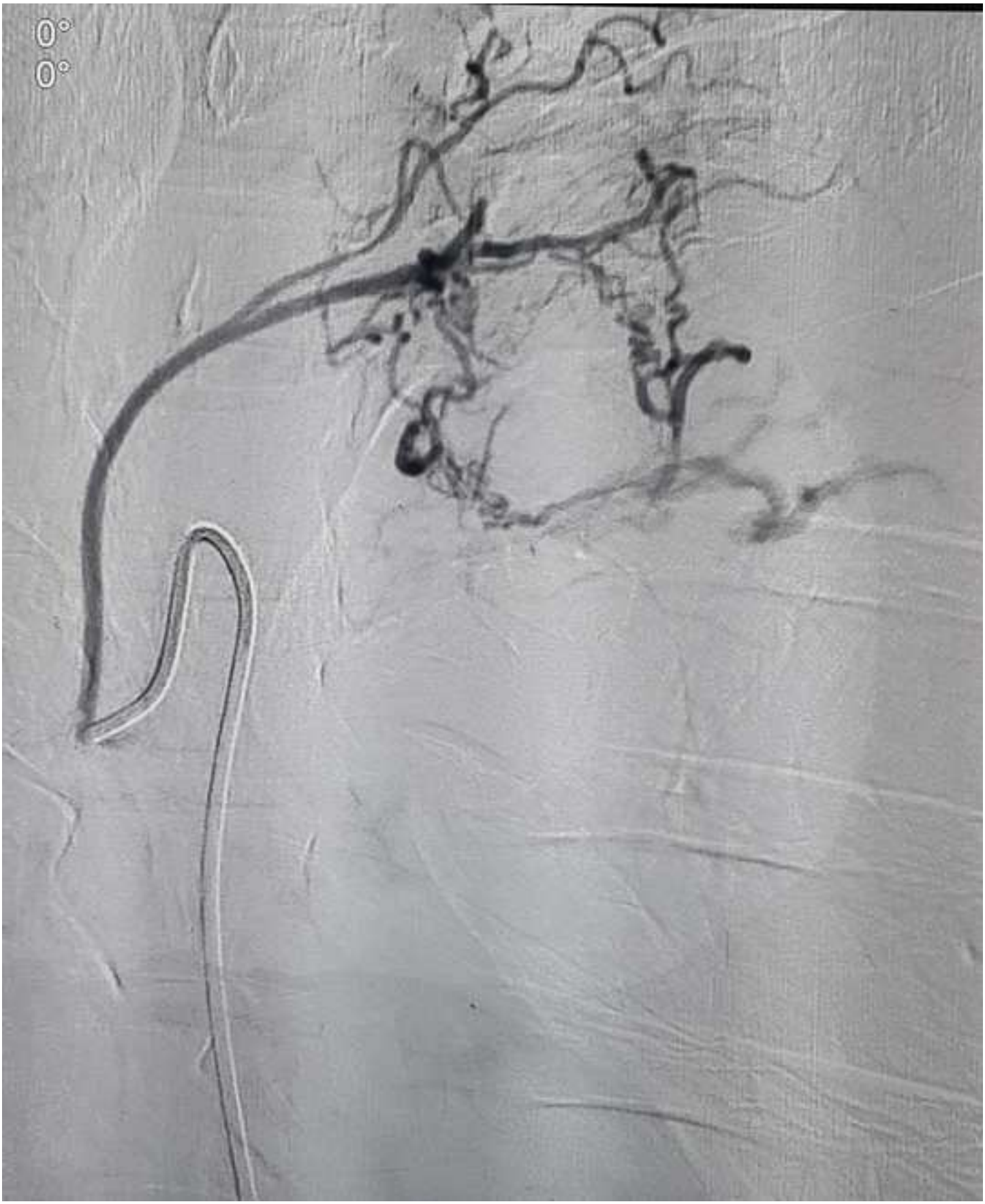

Figure 4A

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Figure 4B

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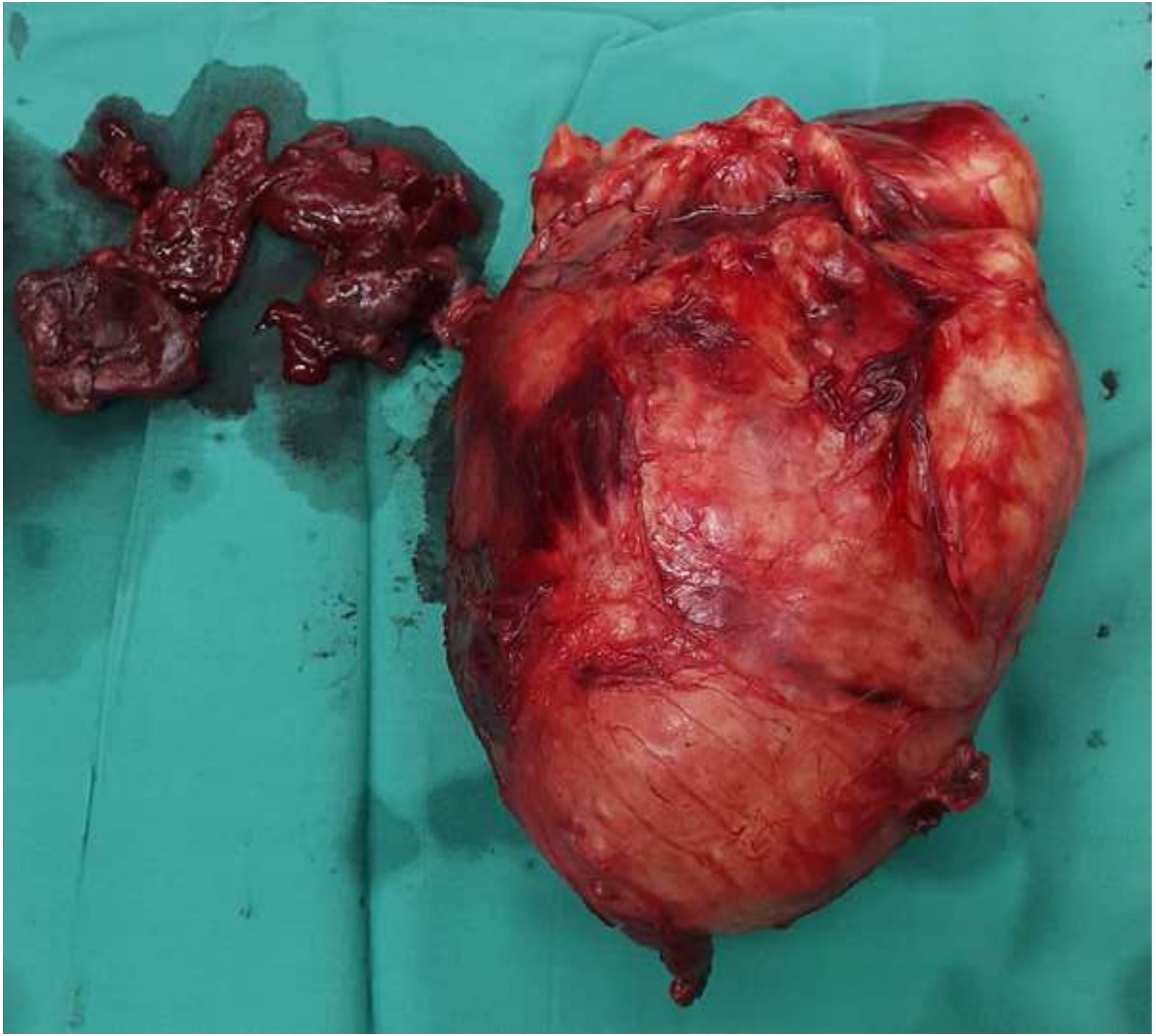

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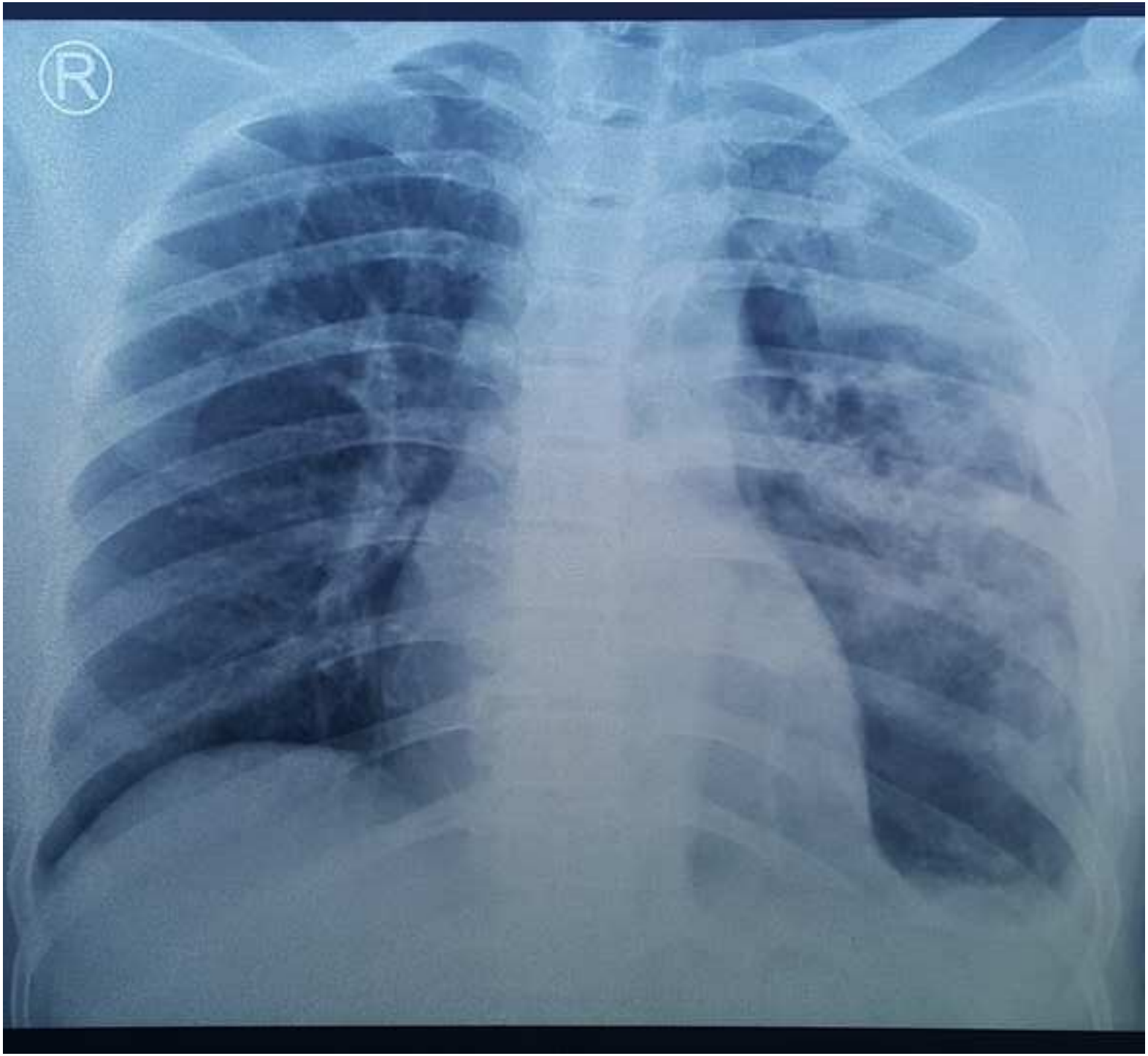

Figure 5B

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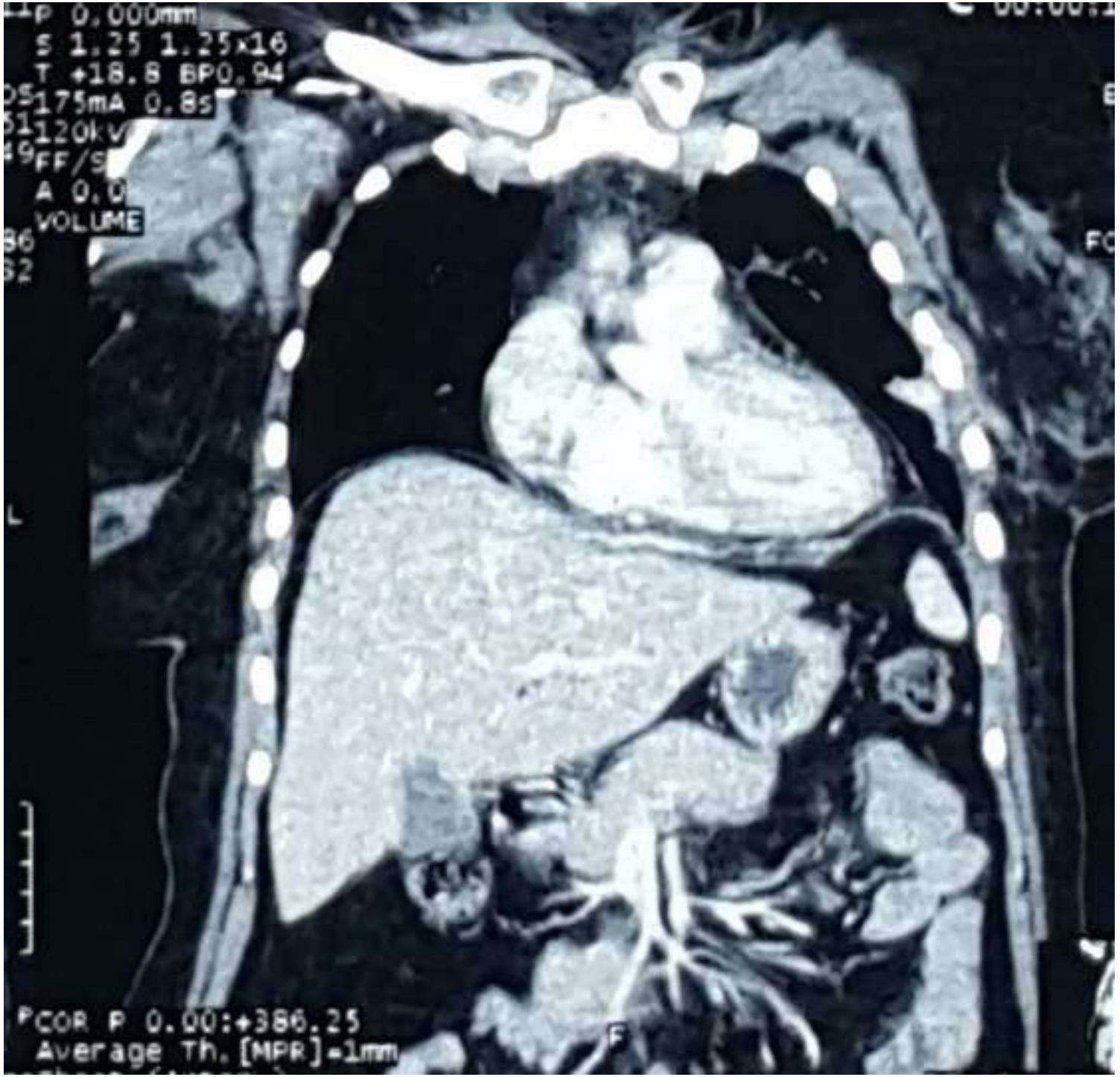

SCARE 2020 Checklist			
Topic	**Item**	**Checklist Item Description**	**Page Number**
Title	**1**	- The words 'case report' should appear in the title. The title should also describe the area of focus (e.g. presentation, patient population, diagnosis, surgical intervention or outcome).	1
Key Words	**2**	- Include three to six keywords that identify what is covered in the case report (e.g. patient population, diagnosis or surgical intervention). - Include 'case report' as one of the keywords.	1
Abstract	**3a**	Introduction and Importance - Describe what is important, unique or educational about the case, and what does this add to the surgical literature.	1
	3b	Case Presentation - Presenting complaints, clinical and demographic details, and the patient's main concerns.	1
	3c	Clinical Findings and Investigations - Clinical findings, investigations performed, main differentials, and subsequent diagnosis.	1
	3d	Interventions and Outcome - Describe the rationale for choosing the intervention. - Describe what was the end result.	1
	3e	Relevance and Impact - Describe the main take-away lessons or potential implications for clinical practice (minimum of three).	1
Introduction	**4**	Background - Describe briefly the area of focus and the relevant background contextual knowledge. Rationale - Describe why the case is different to what is already known and why it is important to report?	1

		- Is the case rare or interesting for the specific healthcare setting, population or country, or is it applicable globally? Guidelines and Literature - Give reference to relevant surgical literature and current standards of care, including any specific guidelines.	
Patient Information	**5a**	Demographic Details - Include de-identified demographic details of the patient (e.g. age, sex, ethnicity, occupation). - Where possible, include other useful pertinent information (e.g. body mass index, hand dominance, income, level of education, marital status).	2
	5b	Presentation - Describe the patient's presenting complaint. - Include a collateral account of the history if relevant. - Describe the patient's mode of presentation (e.g. self-presentation, ambulance or referred by family physician or other hospital clinicians).	2
	5c	Past Medical and Surgical History - Include any previous interventions and relevant outcomes.	2
	5d	Drug History and Allergies - Specify any acute, repeat, and discontinued medications. - Include any allergies and/or adverse reactions.	2
	5e	Family History - Health information regarding first-degree relatives, specifying any inheritable conditions. Social History - Indicate smoking, alcohol, and recreational drug use. - Level of social independence, driving status, and type of accommodation. Review of Systems - If appropriate, report on any other information gathered outside of the focused history.	2

Clinical Findings	**6**	- Describe the general and significant clinical findings based on initial inspection and physical examination.	3
Timeline	**7**	- Summarise the sequence of events leading up to the patient's presentation. - Delays from presentation to diagnosis and/or intervention should be reported. - Use tables or figures to illustrate the timeline of events if needed.	3
Diagnostic Assessment and Interpretation	**8a**	Diagnostic Assessment - Bedside (e.g. urinalysis, electrocardiography, echocardiography). - Laboratory (e.g. biochemistry, haematology, immunology, microbiology, histopathology). - Imaging (e.g. ultrasound, X-ray, CT/MRI/PET). - Invasive (e.g. endoscopy, biopsy).	2
	8b	Diagnostic Challenges - Where applicable, describe what was challenging about the diagnoses (e.g. access, financial, cultural).	2
	8c	Diagnostic Reasoning - Describe the differential diagnoses, why they were considered, and why they were excluded.	2
	8d	Prognostic Characteristics - Include where applicable (e.g. tumour staging).	2
Intervention	**9a**	Pre-Operative Patient Optimisation - Lifestyle (e.g. weight loss). - Medical (e.g. medication review, treating any relevant pre-existing medical concerns). - Procedural (e.g. nil by mouth, enema). - Other (e.g. psychological support).	2
	9b	Surgical Interventions - Describe the type(s) of intervention(s) used (e.g. pharmacological, surgical, physiotherapy, psychological, preventative). - Describe any concurrent treatments (e.g. antibiotics, analgesia, antiemetics, venous thromboembolism	2

		prophylaxis). - Medical devices should have manufacturer and model specifically mentioned.	
	9c	Specific Details regarding Interventions - Describe the rationale behind the treatment offered, how it was performed and time to intervention. - For surgery, include details on the intervention (e.g. anaesthesia, patient position, preparation used, use of other relevant equipment, sutures, devices, surgical stage). - The degree of novelty for a surgical technique/device should be mentioned (e.g. 'first in human'). - For pharmacological therapies, include information on the formulation, dosage, strength, route, and duration.	2-3
	9d	Operator Details and Setting of Intervention - Where applicable, include operator experience and position on the learning curve, prior relevant training, and specialisation (e.g. 'junior trainee with 3 years of surgical specialty training'). - Specify the setting in which the intervention was performed (e.g. district general hospital, major trauma centre).	3
	9e	Deviation from Initial Management Plan - State if there were any changes in the planned intervention(s), and describe these alongside the rationale (e.g. delays to intervention).	3
Follow-Up and Outcomes	**10a**	Specify Details regarding the Follow-Up - When (e.g. how long after discharge, frequency, maximum follow-up length at time of submission). - Where (e.g. home via video consultation, primary care, secondary care). - How (e.g. telephone consultation, clinical examination, blood tests, imaging). - Any specific long-term surveillance requirements (e.g. imaging surveillance of endovascular aneurysm repair	3

		or clinical exam/ultrasound of regional lymph nodes for skin cancer). - Any specific post-operative instructions (e.g. post-operative medications, targeted physiotherapy, psychological therapy).	
	10b	Intervention Adherence and Compliance - Where relevant, detail how well the patient adhered to and tolerated the advice provided (e.g. avoiding heavy lifting for abdominal surgery, or tolerance of chemotherapy and pharmacological agents). - Explain how adherence and tolerance were measured.	3
	10c	Outcomes - Expected versus attained clinical outcome as assessed by the clinician. Reference literature used to inform expected outcomes. - When appropriate, include patient-reported measures (e.g. questionnaires including quality-of-life scales).	3
	10d	Complications and Adverse Events - Precautionary measures taken to prevent complications (e.g. antibiotic or venous thromboembolism prophylaxis). - All complications and adverse or unanticipated events should be described in detail and ideally categorised in accordance with the Clavien-Dindo Classification (e.g. blood loss, length of operative time, wound complications, re-exploration or revision surgery). - If relevant, was the complication reported to the relevant national agency or pharmaceutical company. - Specify the duration of time between completion of the intervention and discharge, and whether this was within the expected timeframe (if not, why not). - Where applicable, the 30-day post-operative and long-term morbidity/mortality may need to be specified. - State if there were no complications or adverse outcomes.	3

Discussion	**11a**	Strengths - Describe the relevant strengths of the case. - Detail any multidisciplinary or cross-speciality relevance.	4
	11b	Weaknesses and Limitations - Describe the relevant weaknesses or limitations of the case. - For novel techniques or devices, outline any contraindications and alternatives, potential risks and possible complications if applied to a larger population.	4
	11c	Relevant Literature - Include a discussion of the relevant literature and, if appropriate, similar published cases. - Describe the implications for clinical practice guidelines and any relevant hypotheses generated.	4
	11d	- Provide a rationale for the conclusions drawn from the case.	4
	11e	Take-Away Lessons - Outline the key clinical lessons from this case report. - Discuss any differences in approach to diagnosis or patient management which the authors might adopt in future similar cases, based on their experience of the case.	4-5
Patient Perspective	**12**	- Where appropriate, the patient should be given the opportunity to share their perspective on the intervention(s) they received (e.g. sharing quotes from a consented and anonymised interview).	N/A
Informed Consent	**13**	- The authors must provide evidence of consent, where applicable, and if requested by the journal. - State the method of consent at the end of the article (e.g. verbal or written). - If not provided by the patient, explain why (e.g. death of patient and consent provided by next of kin). If the patient or family members were untraceable then document the tracing efforts undertaken.	5

Additional Information	**14**	- Please state any author contributions, acknowledgments, conflicts of interest, sources of funding, and where required, institutional review board or ethical committee approval. - Disclose whether the case has been presented at a conference or regional meeting.	5-6
Clinical Images and Videos	**15**	- Where relevant and available, include clinical images to help demonstrate the case pre-, peri-, and post-intervention (e.g. radiological, histopathological, patient photographs, intraoperative images). - Where relevant and available, include a link (e.g. Google Drive, YouTube) to the narrated operative video can be included to highlight specific techniques or operative findings. - Ensure all media files are appropriately captioned and indicate points of interest to allow for easy interpretation.	7
Referencing the Checklist	**16**	- Include reference to the SCARE 2020 publication by stating: ‘This case report has been reported in line with the SCARE Criteria [include citation]’ at the end of the introductory section.	6

Case report

A female with solitary fibrous tumor pleura mimicking pulmonary sclerosing pneumocytoma in low resource setting: A case report

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Abstract

***Background*:** Solitary fibrous tumor of pleura (SFTP) is a rare condition. Clinical symptoms and non-specific radiological features in both tumors make preoperative diagnosis difficult to establish.

***Case presentation*:** A Javanese 47-year-old female complained of chest pain and shortness of breath which an X-ray and CT-Scan of the thorax showed giant cell metastases in the lung. Signs and symptoms indicate pulmonary sclerosing pneumocytoma (PSP) and the patient underwent thoracic surgery which was first performed with angiographic embolization. Furthermore, an anatomical pathology analysis was performed with suspected SFTP, supported by the IHC test, which found CD34 (+), EMA (--), and S100 (--). Thoracic surgery followed by radiotherapy and chemotherapy is recommended in SFTP patients.

***Discussion*:** The SFTP and PSP have non-specific clinical symptoms and radiological features. Anatomic pathology and IHC test are definitive diagnostic tools from SFTP and PSP. Establishing a preoperative diagnosis of SFTP and PSP is quite difficult. Surgical resection is the treatment of choice for both.

***Conclusion*:** The comparison of SFTP and PSP can be seen results of the anatomical pathology analysis and IHC test.

Keywords:

Pulmonary sclerosing pneumocytoma, Solitary fibrous tumors of pleura, Thoracotomy with intercostal incision

Abbreviations

No keyword abbreviations are available

~~1.~~1 Introduction

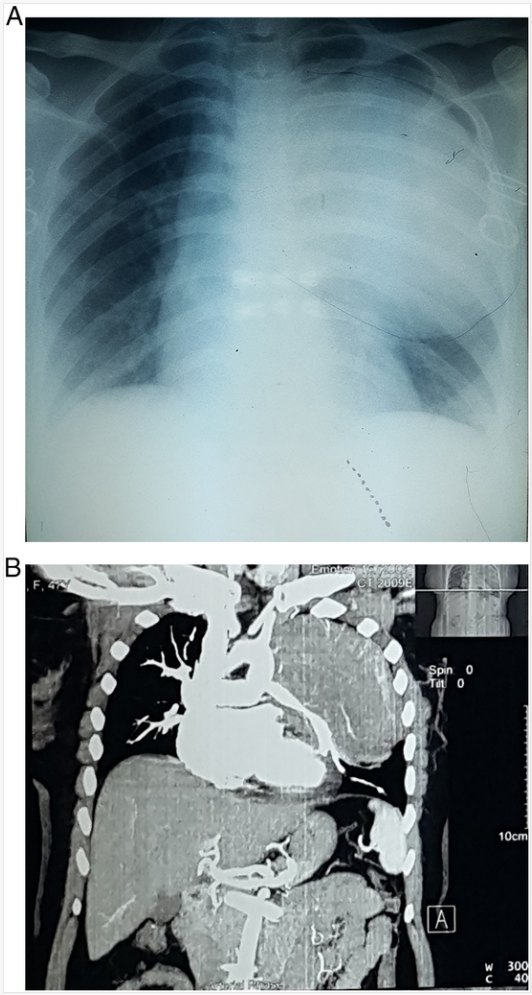
Primary pleural tumors are benign or malignant lesions arising from either the parietal or visceral pleura. The most common of non-mesothelioma primary pleural tumors are solitary fibrous tumors of pleura (SFTP) [1]. In North America, it is estimated that tumor incidence is about 2.8 cases per 100,000 hospitalizations. Malignant mesothelioma is more than 90% of primary pleural tumors. Of the remaining 10%, about 5% are classified as SFTP and the other 5% are various less common primary pleural tumors [2]. Moreover, pleural tumors cause 0.3% to 3.5% thoracic tumors globally [1,3]. The preoperative differential diagnosis that occurs in patients with SFTP is any mass lesion in the chest, ranging from pulmonary carcinoma to various intrapleural sarcomas [4]. In low resource setting, misdiagnosis is possible between SFTP and pulmonary sclerosing pneumocytoma (PSP) [5] because of the similarity of signs and symptoms and limited investigations [6,7]. Based on the description above, we are interested in reporting a case of solitary fibrous tumors of pleura using the surgical case report (SCARE) 2020 guideline [8].

~~2.~~2 Case presentation

A Javanese 47-years-old female complained of chest pain and shortness of breath since 3 months ago. Patients experienced significant weight loss without any decrease in appetite. The patient was suspected of having a lung tumor based on physical and radiological examination (Fig. 1A) when the patient went to the emergency department for asthma exacerbations. Patients were diagnosed with asthma since childhood. Thoracic CT scan with contrast appeared to be a solid lesion (39 HU) which by contrast was increasingly enhanced in the next phase (133 HU). The tumor size of ±10.7 × 14.1 × 9.72 cm in the upper left hemorrhage was attached to the pleura (Fig. 1B). The examination of fiber optic bronchoscopy (FOB) showed a blunt primary carina, narrowing the distal lumen of the main bronchial due to pressure from the posterior wall, narrowing the left main bronchial lumen caused by the pressure of extra lumen mass. The FOB obtained material for anatomic pathology examination and the results showed no malignant cells (Fig. 2).

alt-text: Fig. 1

~~Figure 1.~~Fig. 1



A) X-ray and B) preoperative CT-Scan of the lung.

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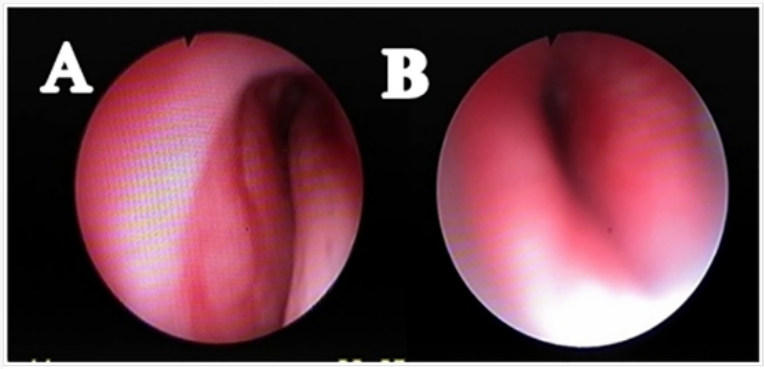
~~Figure 2.~~Fig. 2



A) Right main bronchus and B) left main bronchus.

Embolization was performed, showing the hypervascular mass in the left hemithorax which fed on the left intercostal artery level Th 5–6, the recanalized left bronchial artery, and fine branches of other intercostal artery levels. Post vascularization embolization from the feeding arteries of the superior branches of the left intercostal artery was completely closed. Feeding of the recanalized left bronchial artery and fine branches of other intercostal arteries could not be embolized (Fig. 3).

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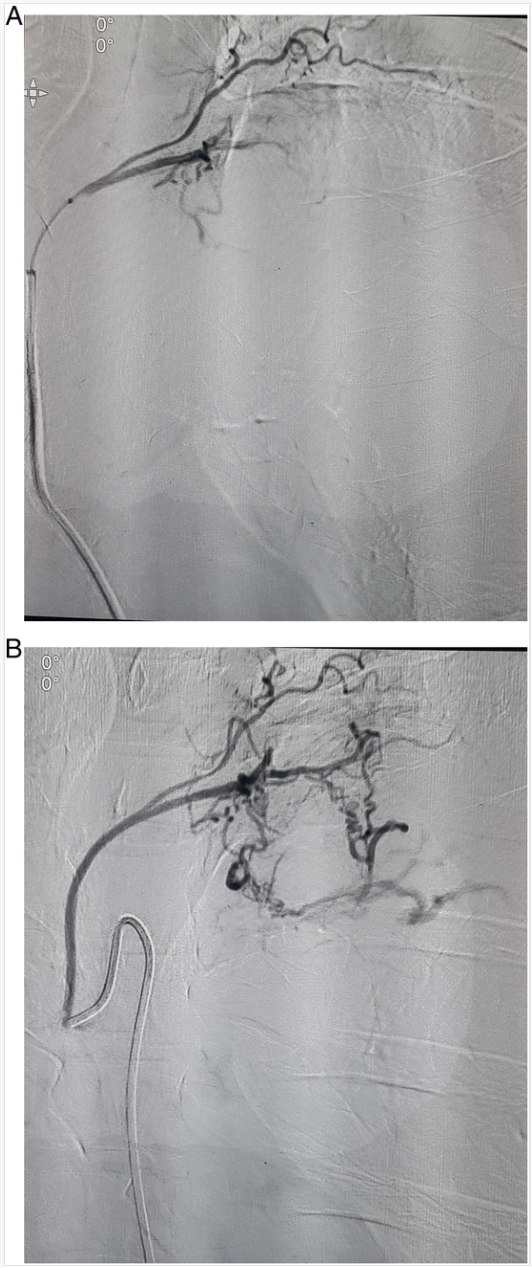
alt-text: Fig. 3

~~Figure 3.~~Fig. 3



Arteriography results recanalized the left bronchial artery.

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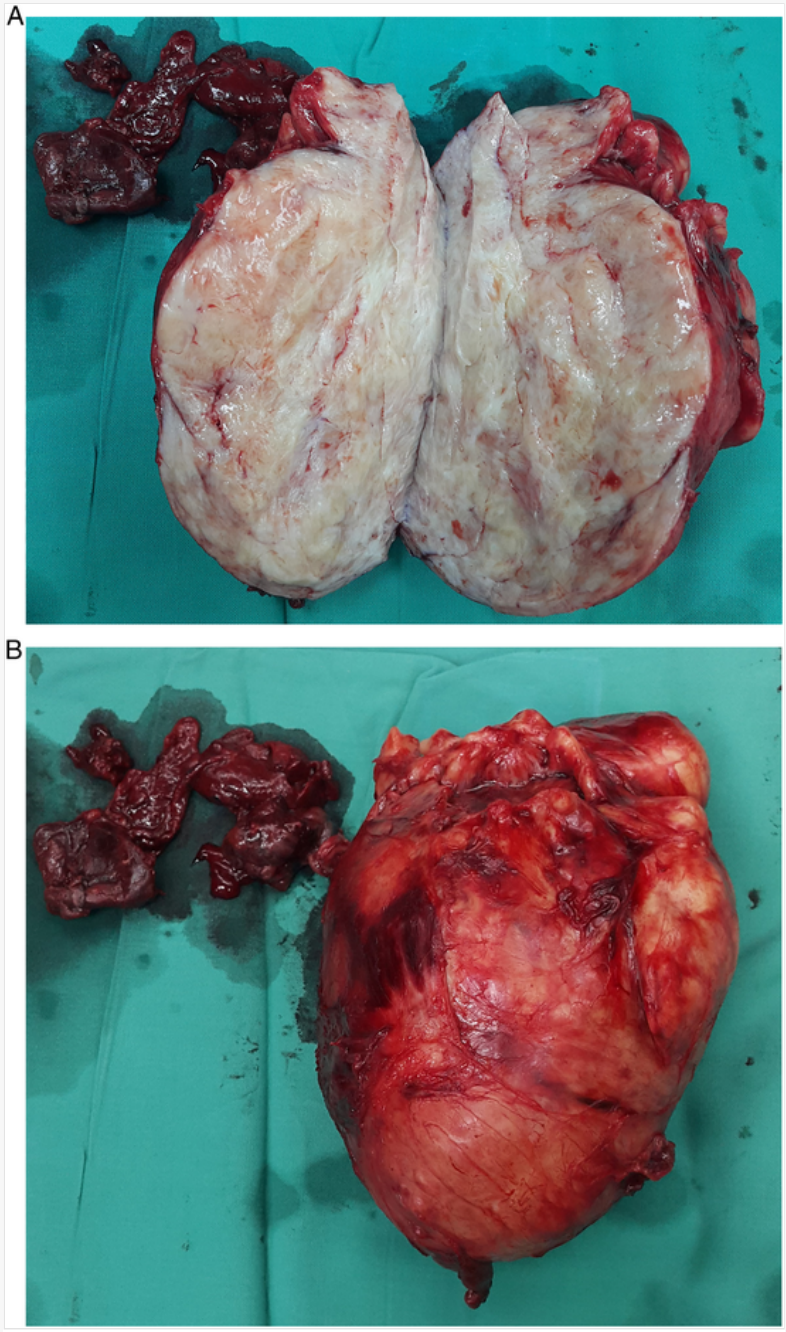
The surgical technique uses an anterolateral thoracotomy approach and enters through the 5th intercostal space. The tumor was seen as a round, well-defined tumor, mounted on the posterior thoracic wall with a size of 12 × 8 × 5 cm (Fig. 4) which anatomical pathology analysis showed tissue sections arranged solidly in irregular patterns, consisting of proliferating oval-spindle-nucleated cells and fine chromatin (SFTP). The results of the immunohistochemical (IHC) test are epithelial membrane antigen (EMA) negative in the membrane and cytoplasm of tumor cells, S100 negative in the cytoplasm of tumor cells, and the cluster of differentiation 34 (CD34) positive in tumor cell membranes. It was concluded that the tumor was a SFTP. A radiological examination was performed to evaluate the patient, showing no mass found in the right/left lung/mediastinum (Fig. 5). After surgery, the patient did not feel any complaints. The patient underwent radiotherapy and chemotherapy. One-year postoperative evaluation, the patient had no respiratory complaints and no clinical or radiological signs of recurrence were found.

alt-text: Fig. 4

~~Figure 4.~~Fig. 4


A
B

Size tumor 12 × 8 × 5 cm.

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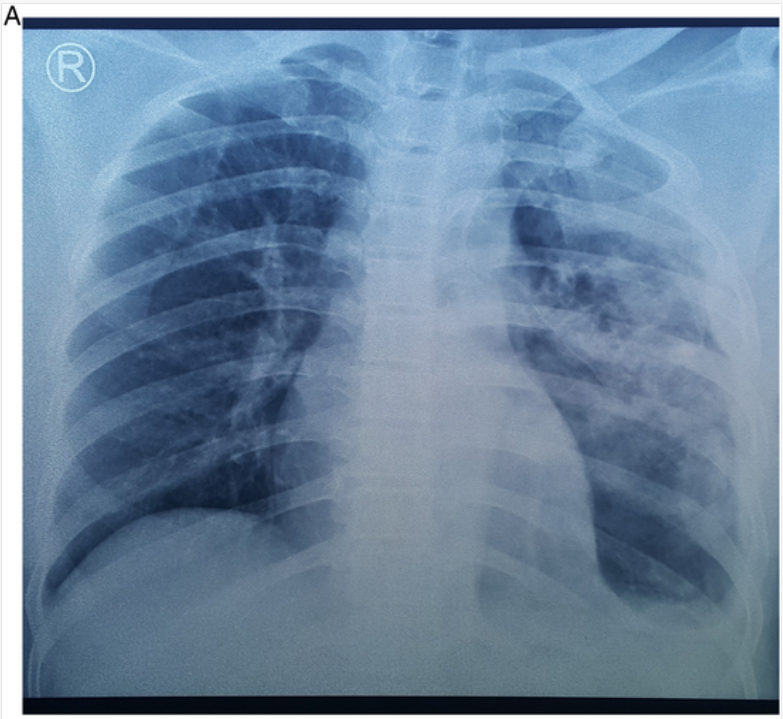
alt-text: Fig. 5

~~**Figure 5.**~~**Fig. 5**

A



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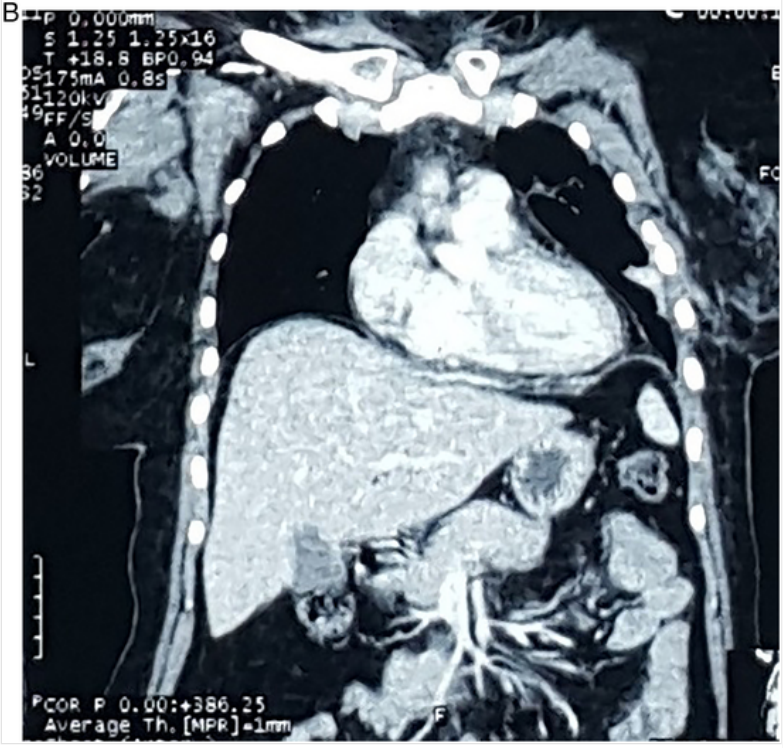
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Postoperative radiological examination.

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~~3.~~3 Discussion

A solitary fibrous pleural tumor is a primary tumor arising from the pleura that can be a benign tumor and has various levels to potentially become malignant [1]. There are two types of primary pleural tumors, namely diffuse and localized. Localized tumors are rare, known as SFTP, which arise from the sub-mesothelial mesenchymal layer. An SFTP occurs in a wide age range (5 to 87 years), often found to occur at the age of the sixth and seventh decades, with quite the same frequency in both sexes [4]. The diagnosis of SFTP and PSP can be distinguished, one of them by using dynamic CT-Scan which dynamic CT-Scan can provide a more accurate picture of the lesion in PSP cases [9]. Dynamic CT-Scan was not performed in our case due to resource constraints. Based on X-ray and CT-scan, giant malignancy is not specific. Support argument diagnosis of SFTP base FOB with malignancy results. The IHC test in SFTP included a primary positive CD34 result, followed by CD99, cytokeratin (S100), and epithelial membrane antigen (EMA) [10]. CD34 is the most frequently used biomarker for SFTP, which in PSP patients the IHC examination was seen for TTF-1, EMA, and pan-cytokeratin immunostaining [11–13].

In cases of SFTP, surgery is the preferred action which in cases of malignancy can be surgical procedures performed safely without a preoperative diagnosis [11]. The surgical procedure chosen was thoracotomy with an intercostal incision which is a surgical technique that is effectively removing the giant tumor in the lung [14]. Before surgery, angiographic embolization is now widely accepted to reduce the occurrence of intraoperative bleeding. Embolization is usually performed 24~~-~~–72 ~~hours~~h before resection to help hemostasis during surgery [15]. Post-surgery, patients are at risk for pain and atelectasis so they need to get special attention for pain management and physiotherapy [16]. Meanwhile, some literature recommends that patients undergo radiotherapy or chemotherapy post-surgery [17,18].

~~4.~~4 Conclusion

A Javanese 47-years-old female complained of chest pain and shortness of breath which an X-ray and CT-Scan of the thorax showed giant cell metastases in the lung. Signs and symptoms indicate PSP and the patient underwent thoracic surgery which was first performed with angiographic embolization. Furthermore, an anatomical pathology analysis was performed with suspected SFTP, supported by the IHC test, which found CD34 (+), EMA (~~-~~–), and S100 (~~-~~–). Thoracic surgery followed by radiotherapy and chemotherapy is recommended in SFTP patients. The comparison of SFTP and PSP can be seen results of the anatomical pathology analysis and IHC test.

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.

Guarantor

Isnin Anang Marhana is the person in charge of the publication of our manuscript.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Sources of funding

None.

Ethical approval

Not applicable.

Research registration

Not applicable.

Cr~~R~~edit~~T~~ authorship contribution statement

Sakina: Data curation, supervision, visualization, investigation, and drafting; **Isnin Anang Marhana:** Conceptualization, methodology, drafting, editing, revising, and reviewing; **Dhihintia Jiwangga ~~Suta Winarno~~:** Methodology, editing, and revising.

Declaration of competing interest

Sakina, Isnin Anang Marhana, and Dhihintia Jiwangga Suta Winarno declare that they have no conflict of interest.

Acknowledgment

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The corrections made in this section will be reviewed and approved by a journal production editor. The newly added/removed references and its citations will be reordered and rearranged by the production team.

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Highlights

Q2

- Solitary fibrous tumor of pleura (SFTP) and pulmonary sclerosing pneumocytoma (PSP) are difficult to distinguish in a low-resource setting.
- SFTP and PSP can be differentiated using an immunohistopathology test (cluster of differentiation 34/CD54 was positive).
- Management of SFTP and PSP are both similar to surgery (thoracotomy with a subcostal incision).

Queries and Answers

Q1

Query: Please review the **given names and surnames** to make sure that we have identified them correctly and that they are presented in the desired order. Carefully verify the spelling of all authors' names as well. If changes are needed, please provide the edits in the author section.

Answer: Yes

Q2

Query: Highlights should only consist of 125 characters per bullet point, including spaces. The highlights provided are too long; please edit them to meet the requirement.

Answer: Done

ISNIN ANANG <isnin.anang@fk.unair.ac.id>

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