



Mature Teratoma of The Lung: A Case Report

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Article Info	Abstract
<p>Article history: Received 16 November 2022 Revised 09 January 2023 Accepted 23 January 2023 Available online 01 February 2023</p> <p>Keywords: Teratoma; lung; tumor; intrapulmonary; germline</p> <p>Correspondence: alphania-r@fk.unair.ac.id</p> <p>How to cite this article: Priangga Adi Wiratama, Vira Yasmina Ramadhani, Alphania Rahniayu. Mature Teratoma of Lung: A Case Report. MAGNA MEDIKA Berk Ilm Kedokt dan Kesehat. 2023; 10(1):109-116</p>	<p>Background: Mature Teratoma is a germinal cell tumor commonly found in the ovaries and testis. Mature Teratoma in the lung region is a rare case. According to the literature, there were only 67 cases worldwide from 1939 to 2007. There was one case in Dr. Soetomo general hospital from January 2013 to December 2017.</p> <p>Objective: Mature Teratoma in the lung region is a rare case. Presenting in a case report would be beneficial for the pathology database</p> <p>Case Presentation: A 19 years old man came to the hospital with shortness of breath three months ago. It was followed by hemoptysis, trichophytic, and coughing out tooth. CT Scan examination showed an irregular walled cavity with semisolid components and calcification and a description of an "air crescent sign." In thoracotomy, tumor excision obtained multilobulated mass size 8.5x6,8x5,6 cm, yellowish-gray white color, and firm consistency. There were also hair and tooth components. The microscopic representation shows pieces of lung tissue with tumor growth coated with squamous epithelia with a stroma containing adnexal skin glands, fatty tissue, cartilage tissue, and serous glands (acinar). <i>Discussion:</i> Mature Teratoma of the lung is one the rare tumors, with symptoms of chest pain, hemoptysis, and trichoptysis. Mature Teratoma of the lung is generally cystic and multiloculated, but can also be solid, encapsulated. On microscopic examination, shows a picture of various components of ectoderm, mesoderm and endoderm.</p> <p>Conclusion: Mature Teratoma of the lung is a rare tumor. Diagnosis was based from clinical findings, radiologic examination, and confirmed with histopathology and immunohistochemistry.</p>

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INTRODUCTION

Mature Teratoma is the most common germ cell tumor of mediastinum but is rarely found in the lungs. Mature Teratoma rarely metastasizes to the lungs. The lesions originate in the third pharyngeal sac and manifest with various clinical and radiological features¹.

Mature Teratoma can be cystic or solid with heterogeneous histopathological features and consist of various components of mature tissue originating from the ectoderm, mesoderm, and endodermis layers². The most common location of teratoma is the sacrococcygeal region, followed by the gonads, ovaries, and testes. However, teratoma can also occur outside the gonads along the mid-line of the body³.

Germinal cell tumors of the lungs occur in adults and children with a variety of age spans

from 10 months to 68 years, primarily diagnosed in the first two decades⁴. Mohr reported the first case of pulmonary Teratoma in 1839¹. There were only 67 reported cases in literature from 1939-2007⁵.

The database of the Anatomical Pathology Laboratory Dr. Soetomo Hospital shows one case of pulmonary Teratoma in the last five years (January 2013-December 2017), which and be discussed in this case report.

CASE PRESENTATION

A 19-year-old man came to Dr. Soetomo Hospital with shortness of breath three months before. The other clinical signs were coughing up blood with hair and teeth. There was no fever. The patient stopped coughing up blood after consuming tranexamic acid.

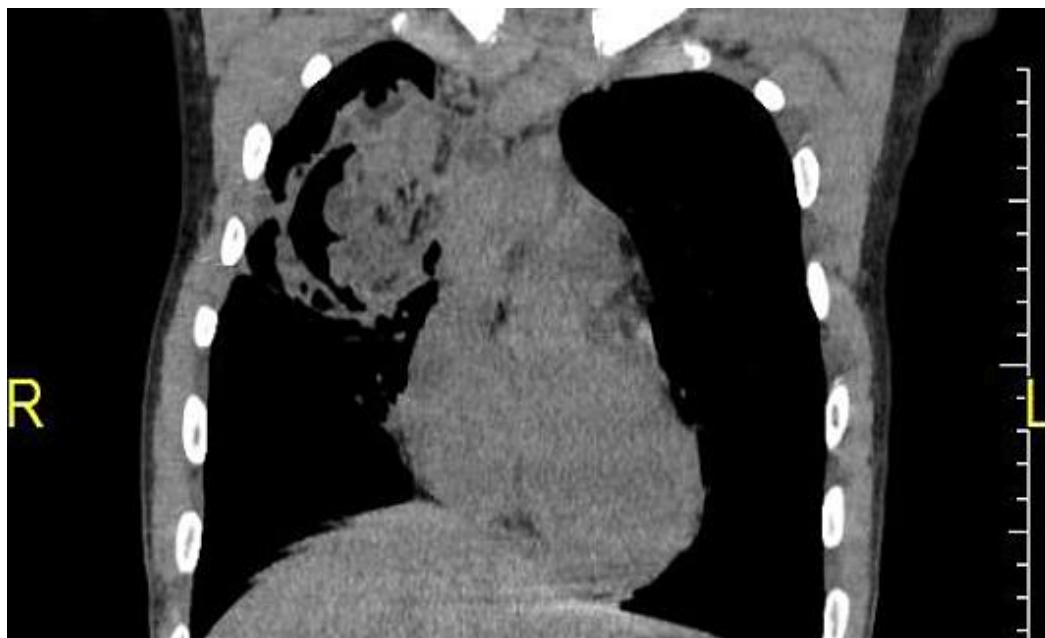


Figure 1. Cystic mass with irregular margin (6,9x7,5x6,8 cm) with semisolid (21-30HU) and calcification (160HU) component with air crescent sign in anterior apical lobe right lung (RSDS Radiology Dept, 2017)

Physical and laboratory examinations showed normal results. Thoracic CT scan with contrast showed a cystic mass of irregular-walled cavity, $\pm 6.9 \times 7.5 \times 6.8$ cm in size with semisolid components (21-30 HU) and calcification (160 HU). The thoracic CT scan also showed an "air crescent sign" in the anterior segment of the superior lobe of the right lung, but without contrast enhancement. CT scan interpretation leads to a picture of a fungus ball accompanied by pulmonary inflammation.

Then, an FNAB (Fine needle aspiration biopsy) examination with CT scan guidance was performed. This procedure using a 25G needle on the mass of the right lung region, which is 6.1×6.0 cm in size and 3.5 cm in depth, in line with thoracic vertebrae V. Microscopic image showed the spread of mature squamous cells, round nuclei, fine chromatin, broad cytoplasm with PMN and mononuclear inflammatory cells. No hyphae. There are no signs of malignancy.

The FNAB examination conclusion is a dermoid cyst or mature teratoma.

Anatomical Pathology Installation received one piece of bilobectomy matter of the lungs' superior lobe tissue. It was 500 grams in weight, $15 \times 6.5 \times 4.5$ cm in size, and the bronchi were not visible. The outer surface was smooth and showed a multilobulated mass of $8.5 \times 6.8 \times 5.6$ cm in size, grey to yellowish colored, and firm in consistency. There were also hair and tooth components.

The microscopic image showed a piece of lung tissue with the growth of a cystic tumor coated with squamous epithelium. The stroma consisted of adnexal glands of the skin, fat tissue, cartilage tissue, and the serous gland's components. Mononuclear inflammatory cells infiltrated the lungs' parenchymal tissue. There were no neuroepithelial or immature components. There was no sign of malignancy. Based on this description, it was concluded as a mature teratoma.

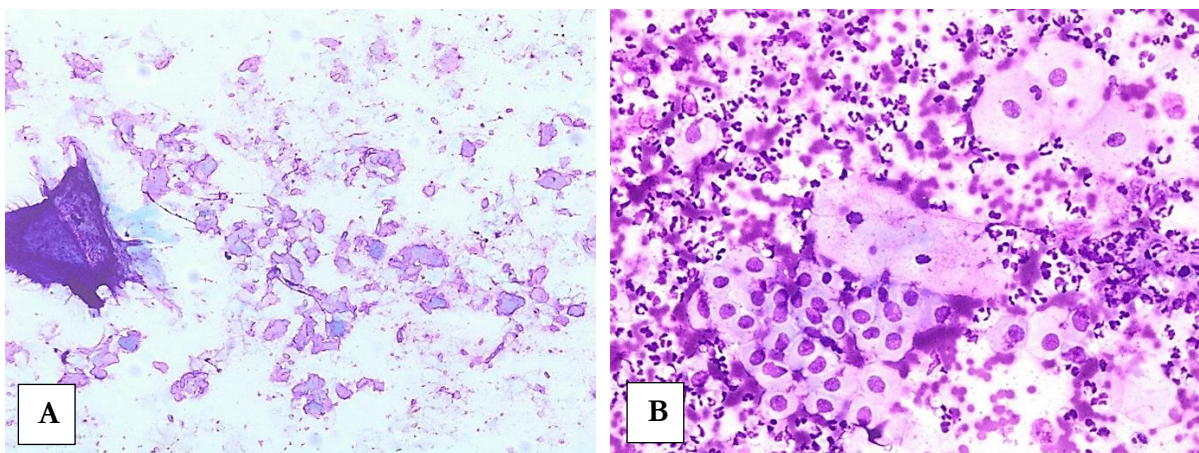


Figure 2. The microscopic FNAB image. A: *Squamae* distribution (Diff-Quik objective 10x) B: Squamous epithelial cells and inflammatory mononuclear cells, PMN, and also RBC (Diff-Quik objective 20x) (RSDS Anatomic Pathology Dept., 2017).

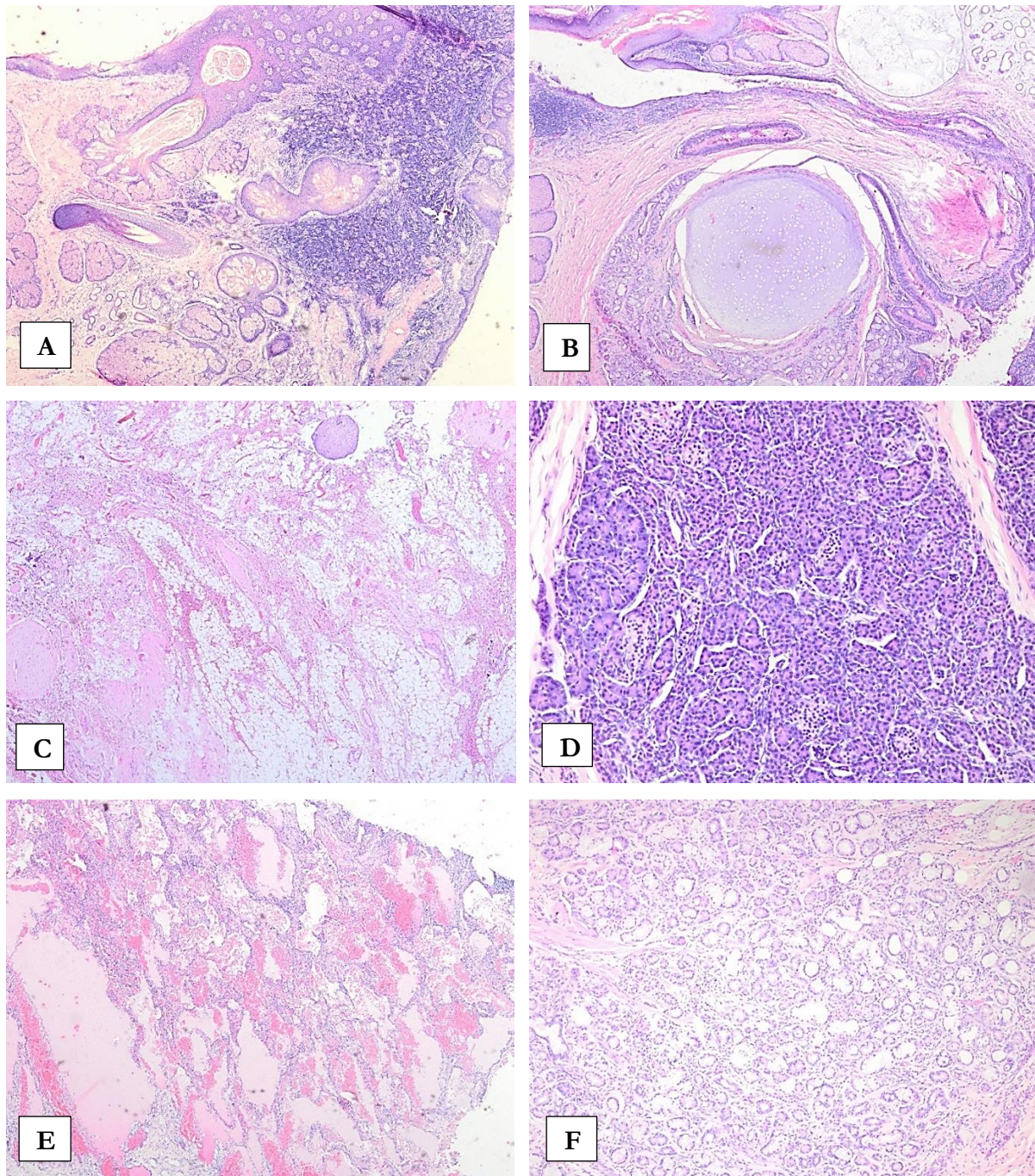


Figure 2. The microscopic image showed various components of mature tissue. A: Squamous epithelia and skin adnexa (Hematoxylin and eosin 10x), B: Mature cartilage bone tissue (Hematoxylin and eosin 10x), C: Adipose tissue (Hematoxylin and eosin 10x), D: Acinus glands (Hematoxylin and eosin 10x), E: Normal alveoli tissue (Hematoxylin and eosin 10x), F: Seromucous glands (Hematoxylin and eosin 10x) (RSDS Anatomic Pathology Dept., 2017).

DISCUSSION

Teratoma is one of the germinal cell tumors originating from the three germinativum cell layers. This tumor is an embryonal neoplasm

that occurs when totipotential germinativum cells are uncontrolled in the development of embryology, then develop into a solid or cystic mass with a diversity of tissue images originating from the three layers of the blasto-

dermis (ectoderm, endodermis, and mesoderm)⁶.

Based on the literature, the most common location of this tumor is in the sacrococcygeal region and gonads, both the ovaries and testis. According to Dr. Soetomo Hospital's pathology database, the most common site of teratoma cases is in the ovaries and intraabdominal.

Teratoma of the pulmonary region is rarely found, and some have been published^{1,5,7,8}. Previous literature stated that intrapulmonary Teratoma was mainly located in the superior lobe (65%), mostly in the anterior segment, for the reason that unidentified yet⁹. That was a similarity in this patient. As far as our knowledge, just one case of Teratoma that involves the entire lobe¹⁰. This tumor's origin is unclear, but it has been suggested that it is anatomically related to the thymus. According to this theory, in early embryos, the primordial teratomatous focus in the thymus may lie in such a position in the potential mediastinum that it is caught up and carried by the respiratory outgrowth from the foregut¹¹. A case report in 2000 confirmed this theory; 15 germinativum cell was found, containing mature thymic tissue, apart from mediastinum¹².

In previous epidemiology studies, most of these teratomas are found in the second to fourth decades (from 10 months to 68 years). This case is in that range. The incidence of this Teratoma is higher in women¹³, a difference in this case report, and it occurs in men. Mature pulmonary Teratoma has clinical features of chest pain (52%), hemoptysis (42%), and cough (39%). The most specific symptom is the discharge of hair components or trichoptysis (13%)⁴. The clinician found some similar complaints in this case.

There is some radiologic challenge in the original diagnosis position of the tumor. In diagnosing mediastinal teratomas, although conventional chest radiographs still play a significant role in the initial evaluation, Computed Tomography (CT) scan is essential for specifying the nature, location, and relationship of the tumor to the surrounding structures¹⁴. Computed Tomography is the best imaging modality for showing tumor morphology and complications. A previous case in India reveals an extension of mediastinal Teratoma causing trichophytic by CT scan¹⁵.

CT scans also reveal tissue density, such as soft tissue, fluid (88%), fat (76%), calcification (53%), and teeth¹⁶, but CT scan result, in this case, lead to an image of a fungus ball accompanied by lung inflammation. This radiologic dilemma also found in the cyst of suspected microorganism in a similar case in 2020¹⁷. In our case, histopathology examination reveals the relationship of the tumor with surrounding tissue to confirm its intrapulmonary position.

Macroscopically the tumor size ranges from 2.8-30 cm. It is generally cystic and multilobulated but can be a solid and encapsulated tumor¹³. The tumor capsule is distinctive and contains various components: teeth, hair, fat, sebum, and cartilage¹⁶. In this case, the tumor was 8.5x6.8x5.6 cm, in the form of a partially cystic multilobulated mass, and obtained hair and tooth components. FNAB features in mature teratomas show a polymorphic cell population consisting of squamous cells, keratin, sebaceous cells, mesenchymal cells, and glandular and columnar epithelial cells¹⁸. In this case, the FNAB showed the distribution of squamae and mature squamous cells, which means FNAB procedure is predictive.

In histopathological examination, mature teratoma shows various components of mature tissue from two or three germinativum layers¹³. In this case, microscopically, we found various components of squamous epithelial cells, adnexal skin glands, fat tissue, cartilage tissue, and serous (acinus) glands. In this case, there were mature tissue components from all layers of germinativum cells, namely the skin coated with squamous epithelium originating from ectodermic, connective tissue, fat tissue, and mature cartilage tissue from the mesodermis, and serous glands (acinous) from endodermis. There is no neuroepithelial component in this patient, so the diagnosis of immature Teratoma can be excluded. The surrounding normal alveoli confirm its intrapulmonary position.

The differential diagnosis of this tumor is pulmonary hamartoma and dermoid cyst. A dermoid cyst is part of mature teratomas, but the difference is that it only consists of ectodermic components without mesodermic and endodermic features. Pulmonary hamartoma is a benign neoplasm consisting of unorganized epithelial and mesenchymal components such as the fibromyxoid stroma, adipose tissue, cartilage, smooth muscle cells, and respiratory epithelium. This tumor arises from fetal embryology but is rarely seen before adulthood. Thoracic lesions appear on chest radiology¹⁹.

Surgery is the chosen treatment for teratoma²⁰. Recurrence is infrequent, so the prognosis is good, except when we find immature components. Even though surgery has been performed, patients still need further therapy with chemotherapy to prevent recurrence¹³.

In summary, the presumption of the rarity of this case also makes it challenging to keep objective in the diagnosis process. Prior radio-

logy findings and then to be confirmed by histopathology, led us to state it as lung teratoma, a rare case clearly.

CONCLUSION

Pulmonary mature Teratoma is a rare tumor. A 19-year-old male patient came with clinical signs of hemoptysis, trichoptysis, and coughing out teeth. The CT scan showed irregular-walled cavities with semisolid and calcified components and a "air crescent sign" on the right lung. A multilobulated mass was obtained on macroscopic examination, and there were hair and tooth components. Microscopic examination found various kinds of ectodermis, mesodermis, and endodermis components. In this case, pulmonary mature teratoma diagnosis is based on anamnesis, physical examination, imaging modalities, and histopathology. Therefore, a good multidisciplinary team is needed between pulmonary specialists, thoracic and cardiovascular surgery, radiology, and anatomical pathology for optimal management of pulmonary teratoma. At present, the preferred treatment for lung maturation teratoma is surgery.

REFERENCES

1. Saini ML, Krishnamurthy S, Kumar R V. Intrapulmonary mature Teratoma. *Diagn Pathol* [Internet]. 2006 [cited 2022 Sep 3]; 1(1): 38. Available from: /pmc/articles/PMC1626485/
2. Serraj M, Lakranbi M, Ghalimi J, Ouadnoui Y, Smahi M. Mediastinal mature teratoma with complex rupture into the lung, bronchus and skin: a case report. *World J Surg Oncol* [Internet].

- 2013 Jun 1 [cited 2022 Sep 3]; 11:125. Available from: [/pmc/articles/PMC3674925/](#)
3. Kalyoussef E, Rosen A, Tomovic S. Benign cystic Teratoma of the parotid gland. *Laryngoscope*. 2011;121(SUPPL. 4):S129.
 4. Mondal SK, DasGupta S. Mature cystic teratoma of the lung. *Singapore Med J*. 2012;53(11):237–9.
 5. Vigg A, Khulbey SK, Kumar Agarwal S, Dikshit V, Sathpathy A, Srinivas U, et al. Intra-pulmonary Teratoma: A Rare Case Radiology Forum Figure 1. Chest radiograph (posterior-anterior view) showing a well-defined opacity in the right lower zone.
 6. Chakravarti A, Shashidhar TB, Naglot S, Sahni JK. Head and Neck Teratomas in Children: A Case Series. *Indian J Otolaryngol Head Neck Surg* [Internet]. 2011 [cited 2022 Sep 3]; 63(2): 193. Available from: [/pmc/articles/PMC3102158/](#)
 7. Dar RA, Mushtaque M, Wani SH, Malik RA. Giant Intrapulmonary Teratoma: A Rare Case. *Case Rep Pulmonol*. 2011; 2011:1–3.
 8. Sawant AC, Kandra A, Narra SR. Rare disease: Intrapulmonary cystic Teratoma mimicking malignant pulmonary neoplasm. *BMJ Case Rep* [Internet]. 2012 [cited 2022 Sep 3]; 2012. Available from: [/pmc/articles/PMC3433504/](#)
 9. Bawazir AA, Alrossais NM, BinSaleh Y, Alamodi A, Alshammari A. A Case Report of Intrapulmonary Teratoma in the Right Upper Lung Zone in a 35-year-old Female Patient. *Cureus* [Internet]. 2019 Jan 8 [cited 2022 Sep 8]; 11(1). Available from: [/pmc/articles/PMC6411339/](#)
 10. Giunchi F, Segura JJ. Primary malignant Teratoma of lung: Report of a case and review of the literature. *Int J Surg Pathol*. 2012;20(5):523–7.
 11. Day DW, Taylor SA. An intrapulmonary teratoma associated with thymic tissue. *Thorax* [Internet]. 1975 [cited 2022 Sep 8];30(5):582–7. Available from: <https://pubmed.ncbi.nlm.nih.gov/1198403/>
 12. Asano S, Hoshikawa Y, Yamane Y, Ikeda M, Wakasa H. An intrapulmonary teratoma associated with bronchiectasia containing various kinds of primordium: a case report and review of the literature. *Virchows Arch* 2000 4364 [Internet]. 2000 [cited 2022 Sep 8]; 436(4): 384–8. Available from: <https://link.springer.com/article/10.1007/s004280050463>
 13. Travis WD, Brambilla E, Nicholson AG, Yatabe Y, Austin JHM, Beasley MB, et al. WHO Classification of Tumours of the Lung, Pleura, Thymus and Heart. *J Thorac Oncol* [Internet]. 2015 [cited 2022 Sep 3];10(9): 1243–60. Available from: http://content.wkhealth.com/linkback/openurl?sid=WKP_TLP:landingpage&an=01243894-900000000-98927%5Cnhttp://www.ncbi.nlm.nih.gov/pubmed/26291008
 14. Awad AK, Fatima A, Khurana S, Elbadawy MA, Elseidy SA. Right upper lobe intrapulmonary mature cystic teratoma. An unusual location with unusual associations and a review of the literature. *Int J Surg Case Rep*. 2022 Jan 1;90:106683.
 15. Bachh AA, Haq I, Gupta R, Boinapally RM, Sudhakar S. Benign mediastinal

- teratoma with intrapulmonary extension presenting with trichoptysis. *Respir Med CME*. 2010 Jan 1;3(3):189–91.
16. Zisis C, Rontogianni D, Stratakos G, Voutetakis K, Skevis K, Argiriou M, et al. Teratoma occupying the left hemithorax. *World J Surg Oncol* [Internet]. 2005 Nov 22 [cited 2022 Sep 3];3:76. Available from: [/pmc/articles/PMC1308873/](#)
 17. Mardani P, Naseri R, Amirian A, Shahriarirad R, Anbardar MH, Fouladi D, et al. Intrapulmonary mature cystic Teratoma of the lung: case report of a rare entity. *BMC Surg* [Internet]. 2020 Sep 14 [cited 2022 Sep 8]; 20(1): 1–6. Available from: <https://bmcsurg.biomedcentral.com/articles/10.1186/s12893-020-00864-y>
 18. Siddiqui FA, Jain A, Maheshwari V, Beg MH. FNA diagnosis of teratoma lung: A case report. *Diagn Cytopathol* [Internet]. 2010 Oct 1 [cited 2022 Sep 3]; 38(10): 758–60. Available from: <https://onlinelibrary.wiley.com/doi/full/10.1002/dc.21318>
 19. Umashankar T, Devadas AK, Ravichandra G, Yaranal P. Pulmonary hamartoma: Cytological study of a case and literature review. *J Cytol* [Internet]. 2012 Oct 1 [cited 2022 Sep 3]; 29(4): 261. Available from: <https://www.jcytol.org/article.asp?issn=0970-9371;year=2012;volume=29;issue=4;spage=261;epage=263;aulast=Umashankar>
 20. Hammen I, Lal Yadav A. Teratoma as unusual cause of chest pain, hemoptysis and dyspnea in a young patient. *Respir Med Case Reports*. 2018 Jan 1;23:77–9.