

## Pulmonary Papillary Proliferation Within a Mature Cystic Teratoma of the Ovary: A Rare Case Report and Review of Literature

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### ABSTRACT

**Background:** Mature cystic teratomas are the most common germ cell tumors of the ovary. Both malignant and benign tumors may arise inside mature teratomas. Only 42 cases of pulmonary papillary adenomas are described in the literature, with only two cases of pulmonary papillary adenoma inside a mature cystic teratoma of the ovary.

**Case Report:** This case report highlights the presence of a pulmonary papillary adenoma in a mature cystic teratoma of the ovary in a 28-year-old woman, which is the third case documented in the literature. This report also includes a review of the literature discussing its histomorphological and immunohistochemical features.

**Conclusion:** Pulmonary papillary adenomas within ovarian mature cystic teratomas represent an exceedingly rare phenomenon warranting further research.

**Keywords:** Pulmonary papillary proliferation, ovary, mature teratoma.



#### Cite this article as:

Yalçın AC, Yalçın N, Ayazoğlu MS, Fındık Güvendi G, Bedir R. Pulmonary Papillary Proliferation Within a Mature Cystic Teratoma of the Ovary: A Rare Case Report and Review of Literature. J Clin Pract Res 2024;46(4):399–401.

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**Submitted:** 22.02.2024  
**Revised:** 16.05.2024  
**Accepted:** 24.05.2024  
**Available Online:** 23.08.2024

Erciyes University Faculty of  
Medicine Publications -  
Available online at [www.jcprres.com](http://www.jcprres.com)

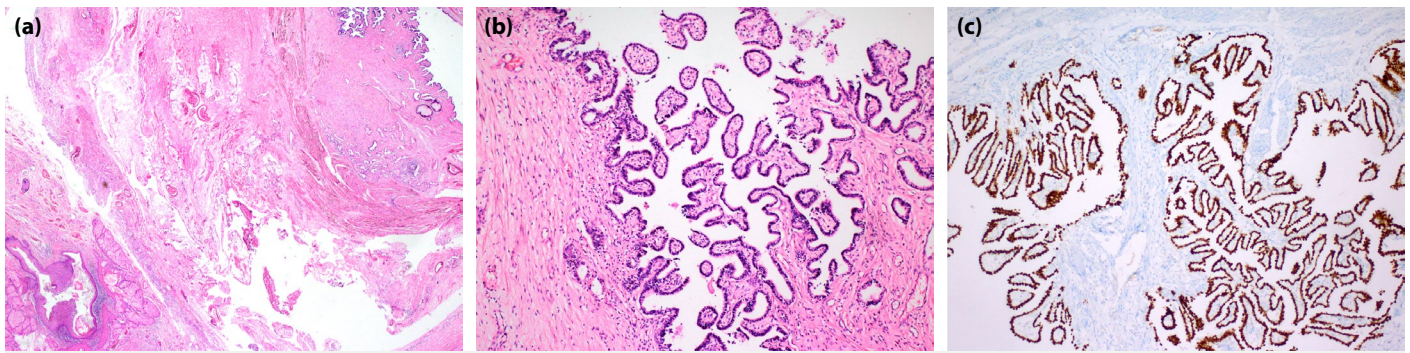


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### INTRODUCTION

Mature cystic teratomas, which encompass elements derived from all three germ layers, are the most common germ cell tumors of the ovary. These tumors, predominantly seen in young women between ages 20 and 40, comprise 20–25% of all ovarian tumors. The most common complications of these tumors are torsion and rupture, but their potential for malignant transformation, albeit rare (1–2%), necessitates thorough evaluation. The most common malignant transformations are to squamous cell carcinoma, followed by thyroid carcinomas.<sup>1</sup>

Pulmonary papillary adenomas are benign lesions of the lung. There are only 42 reported cases of this tumor, and this lack of knowledge makes them hard to diagnose.<sup>2</sup> These tumors are mostly located in the periphery of the lungs. They are composed of cells similar to Clara cells and type II pneumocytes and are thought to arise from pluripotent cells of the bronchioalveolar epithelium, with the potential for malignant transformation.<sup>3</sup> There are only two cases of pulmonary papillary adenoma inside a mature cystic teratoma of the ovary in the literature, and our case report will be the third.<sup>4,5</sup>



**Figure 1.** (a) Papillary neoplasm within a mature teratoma, surrounded by skin and skin appendages (hematoxylin and eosin, x20). (b) Papillae with fibrovascular cores covered by cuboidal epithelium, featuring oval-circular nuclei and eosinophilic cytoplasm (H&E, x200). (c) Diffuse positive staining with thyroid transcription factor-1 (TTF-1) in the papillary neoplasm (x400).

## CASE REPORT

A 28-year-old female patient, previously managed at a different hospital, presented to our gynecology and oncology department with her magnetic resonance imaging (MRI) report. The MRI report displayed a 55 x 46 mm sized “dermoid cyst” in the right ovary. Further evaluation revealed no history of malignancy in herself or her family. She had negative tumor markers and no other masses in additional imaging studies. Because of her desire to preserve fertility, a right ovarian cystectomy was performed.

Macroscopically, evaluation revealed a cystic mass sized 3 x 2.2 x 1 cm, containing hair and sebaceous material inside. Microscopically, the tumor consisted of skin, skin appendages, and gastric and colonic mucosa. In one field, a distinct tumor characterized by cuboidal cells featuring oval-circular nuclei, eosinophilic cytoplasm overlaying papillary structures with anastomosing fibrovascular cores was observed (Fig. 1a, b). Pleomorphism, atypia, necrosis, and mitosis were absent. Even though the glass-ground appearance and other nuclear features of papillary thyroid carcinoma were absent, a differential diagnosis was required since papillary thyroid carcinoma is a more common tumor in mature cystic teratomas than the extremely rare papillary adenoma of the lung. The immunohistochemistry studies revealed thyroid transcription factor-1 (TTF-1) positivity with negative paired box gene 8 (PAX8) and thyroglobulin, which dismissed thyroïdal origin (Fig. 1c). To differentiate from choroid plexus papilloma, S100 and Glial fibrillary acidic protein (GFAP) stains were used. The absence of positive staining with S100 and GFAP excluded this diagnosis. The tumor showed diffuse positivity with the epithelial marker cytokeratin 7 and a low Ki-67 proliferation index (1%). With all these findings, the case was diagnosed as pulmonary papillary adenoma.

## DISCUSSION

Pulmonary papillary adenoma is a benign tumor of the lung. These tumors arise from cuboidal cells that have features of type II pneumocytes and Clara cells. There are less than 50 cases in the literature, making this a challenging diagnosis. Furthermore, these tumors may arise in mature cystic teratomas with significant resemblance to some malignant tumors.<sup>2</sup>

The first case reported in the literature by Damiani depicted a lesion consistent with a cystic tumor without papillary structures and accompanying Brenner tumor.<sup>4</sup>

The second case was from Lane and colleagues, which had the classical nuclear features of thyroid papillary carcinoma.<sup>5</sup> In this lesion, surfactant positivity with thyroglobulin negativity excluded thyroïdal origin, aiding the differential diagnosis.

Thyroid tissue is a common component of mature cystic teratomas and may also appear as “struma ovarii.” Because of this relationship, the most common papillary neoplasia arising inside mature cystic teratomas are thyroid papillary carcinomas. In our case, the typical nuclear features of papillary thyroid carcinoma were absent. There was no normal thyroid tissue around the lesion. Additionally, TTF-1 positivity with thyroglobulin and PAX8 negativity supported the exclusion of a papillary thyroid carcinoma diagnosis.

In our case, the second possible diagnostic resemblance was normal choroid plexus and choroid plexus papilloma. To exclude this diagnosis, S100 and GFAP staining were performed, which did not reveal positive staining. The absence of normal glial tissue around the lesion also helped exclude this diagnosis.

The pathogenesis of papillary adenomas is debatable.<sup>2</sup> The existence of a complex architectural structure raises concern for primary lung adenocarcinoma. The morphological features of our case excluded malignancy, but even benign tumors may be misdiagnosed as primary well-differentiated adenocarcinoma of the lungs. In our case, there was no pleomorphism, atypia, mitosis, or necrosis. The Ki-67 proliferation index was low (1%). The systemic evaluation did not reveal any primary or metastatic lesions. These findings collectively contributed to differentiation from malignancy.

## CONCLUSION

Pulmonary papillary adenomas within ovarian mature cystic teratomas represent an exceedingly rare phenomenon warranting further research. The differential diagnosis of papillary thyroid carcinoma, normal choroid plexus, and choroid plexus papilloma is required. After the exclusion of these lesions, pathologists should be aware of this extremely rare diagnosis.

**Informed Consent:** Written informed consent was obtained from patients who participated in this study.

**Author Contributions:** Concept – ACY, NY, RB; Design – RB, ACY; Supervision – RB; Resource – MSA, GFG; Materials – MSA, GFG; Data Collection and/or Processing – ACY, NY, RB; Analysis and/or Interpretation – RB; Literature Search – ACY, NY, RB; Writing – ACY; Critical Reviews – RB.

**Conflict of Interest:** The authors have no conflict of interest to declare.

**Use of AI for Writing Assistance:** Not declared.

**Financial Disclosure:** The authors declared that this study has received no financial support.

**Peer-review:** Externally peer-reviewed.

## REFERENCES

1. Balık G, Ustüner I, Bedir R, Ural UM, Kağıtçı M, Güven ES. Appendix and uterus metastasis of squamous cell carcinoma arising from mature cystic teratoma of the ovary. *Case Rep Obstet Gynecol* 2013; 2013: 474891. [\[CrossRef\]](#)
2. Liu P, Feng J, Yang M, Chen J, Fu L, Lu J. Pulmonary papillary adenoma with malignant potential: a case report and literature review. *Diagn Pathol* 2022; 17(1): 81. [\[CrossRef\]](#)
3. Ma H, Wang Y, Chen P, Zhang Z, Xu J. Pulmonary papillary adenoma with malignant transformation: report of one case and review of the literature. *Int J Clin Exp Pathol* 2020; 13(4): 792–8.
4. Damiani S. Pulmonary papillary adenoma-like tumour arising in ovarian teratoma. *Virchows Arch* 2004; 445(1): 96–7. [\[CrossRef\]](#)
5. Lane S, Newman A, Vergine M, Allan K, Usifo F, Yeoh CC, et al. Pulmonary-derived papillary adenomatoid proliferation arising in ovarian mature cystic teratoma. *Clin Exp Obstet Gynecol* 2020; 47(4): 611–3. [\[CrossRef\]](#)