

Pulmonary-derived papillary adenomatoid proliferation arising in ovarian mature cystic teratoma

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Summary

Introduction: Primary lung papillary neoplasms are very rare and less than 30 cases of papillary adenomas have been described. Lung-derived papillary lesions arising in ovarian mature cystic teratoma are extremely rare and, to the best of the authors' knowledge, only one case has been reported. Herein they report the second case and discuss the differential diagnoses. **Case Report:** A 32-year-old woman presented with bilateral ovarian mature cystic teratoma (MCT) ("dermoid"). The right side teratoma, very close to the thyroid tissue (struma ovarii), presented a morphologically benign mass, with complex papillary structures and psammoma bodies, which mimicked a thyroid papillary carcinoma (TPC). Immunohistochemistry showed diffuse positive staining with EMA, cytokeratins, and patchy positive staining with TTF1 and surfactant apoprotein A. Thyroglobulin, PAX-8, HMBE1, chromogranin, synaptophysin, CD56, CEA, p63, and BRAF were negative. The work-up for metastasis was negative. Close clinical and instrumental follow-up was decided following consultation with the lung and gynaecological multidisciplinary team. **Conclusion:** This is the second case of benign appearing, lung-derived papillary neoplasm arising in an ovarian MCT. In view of the clinical context, histological criteria used in lung pathology cannot be applied with confidence in this case. The most appropriate term would be pulmonary-derived papillary adenomatoid proliferation. This novel entity will be useful for pathologists and clinicians in diagnosing and differentiating this very rare disease from other lesions arising in ovarian teratomas.

Key words: Lung; Ovary; Teratoma; Papillary; Adenoma.

Introduction

Mature cystic teratoma (MCT) is the commonest ovarian tumour and recurs during the reproductive age in > 80% of cases. MCT is composed of adult-type tissue usually representing all three germ layers. However, microscopic foci of fetal-type tissue can also be found in many otherwise typical MCT. Malignant transformation of the somatic component in MCT is rare and squamous cell carcinoma is the most common subtype followed by thyroid carcinoma, adenosquamous carcinoma, intestinaltype adenocarcinoma, and neuroendocrine tumours [1].

Lung-derived papillary lesions are extremely rare and, therefore, understudied. The differential diagnosis of papillary tumour arising in MCT can be arduous. In addition, the histopathological criteria used in lung pathology to distinguish benign lesions from malignant counterparts, due to the teratomatous background, cannot be used. Until now, only one case of papillary lesion arising in MCT has been documented [2]. The authors report the second case, which morphologically mimicked thyroid papillary carcinoma (PTC), and discuss the differential diagnoses.

Case Report

A 32-year-old woman presented with bilateral ovarian complex masses. Ultrasonography showed features consistent with cystic teratoma ("dermoid"). The family history was negative for ovarian and breast cancer. Bilateral salpingo-oophorectomy was performed. On macroscopic examination, both ovaries showed the conventional features of MCT. The right side neoplasm was intact and measured 200 × 145 × 110 mm. The contralateral tumour was received incised and collapsed and measured 45 × 35 × 10 mm. Histologically, both masses presented typical features of MCT. The left side showed no worrisome features, while the right side, within a cystic cavity lined by respiratory-type epithelium and very close to the thyroid tissue (struma ovarii), presented an 8 mm mass with complex papillary structures with psammoma bodies (Figure 1). Some papillae contained foamy macrophages and papillae with fibrohyaline cores were easily identified. The papillae contained a fibro-vascular core and were covered by cuboidal/columnar epithelium with nuclear overlapping, clearing, and grooving (Figure 2). No definite intranuclear inclusions were seen. The mitoses were extremely rare, no necrosis was present and there was no infiltration of the adjacent teratomatous tissue. The tumour was organ confined.

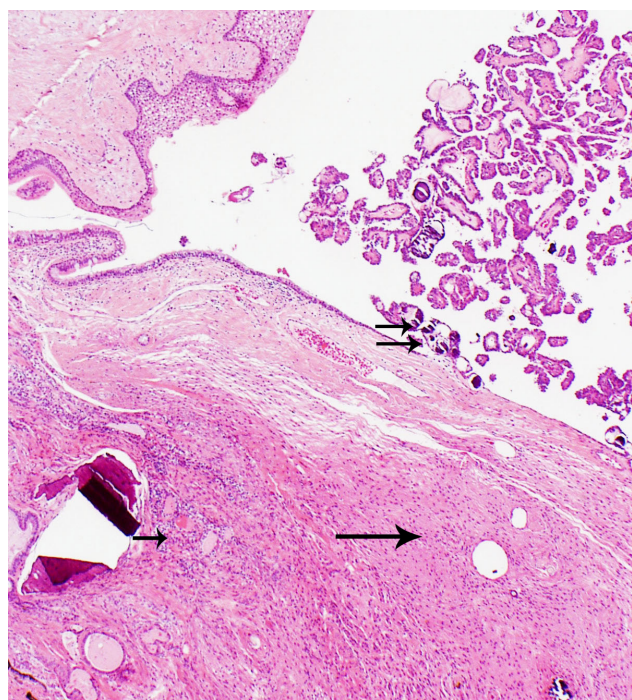


Figure 1. — A papillary neoplasm, with psammoma bodies (double arrows), arising within a cystic cavity lined partly by respiratory-type epithelium and partly by squamous epithelium. Mature neural tissue (long arrow) intermingled with thyroid tissue (short arrow) are present.

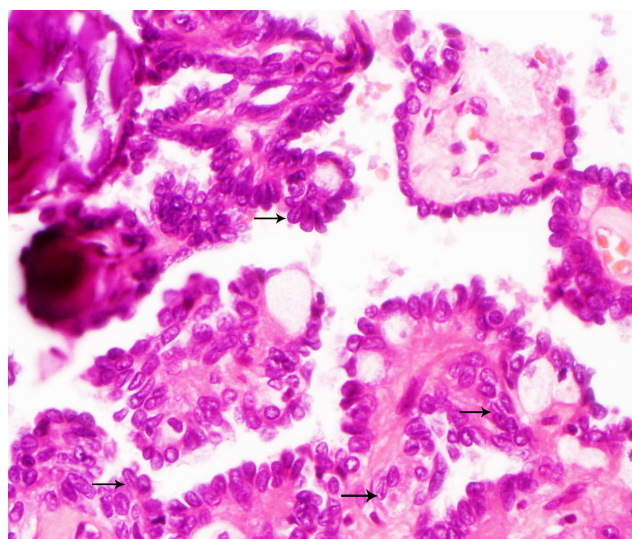


Figure 2. — Papillae with fibro-vascular cores covered by cuboid epithelium with nuclear overlapping, clearing, and grooving (arrows). In the left corner, psammoma bodies are identified.

Immunohistochemistry showed diffuse positive staining with EMA, cytokeratins (AE1/3), cytokeratin 7, cytokeratin 19, and patchy positive staining with TTF1 and surfactant apoprotein A (Figure 3A, B). Thyroglobulin, PAX-8, HMBE1, chromogranin, synaptophysin, CD56, CEA, p63,

and BRAF were negative. Ki67 showed < 1% of nuclear staining.

The work-up for metastasis was negative and after consultation in the lung and gynaecological multidisciplinary team, close clinical and instrumental follow-up was decided.

Discussion

The authors report a case of lung-derived papillary lesion arising in an otherwise benign MCT. To the best of their knowledge, this is the second case of a lung-originated papillary neoplasm, arising in the context of an ovarian MCT. The first case was reported by Damiani [2], however, the microscopic photographs of this case depict a benign-appearing lesion, with no particularly complex papillary structures, in the context of a benign Brenner tumour with mucinous metaplasia. Interestingly, in the case reported herein, the lesion was very close to the struma ovarii, which, in addition to the classic nuclear features of PTC, might easily create a diagnostic pitfall. Careful examination showed that the lesion arose in a cystic cavity lined by respiratory-type epithelium. Immunohistochemistry showed positive staining for surfactant apoprotein A and TTF1 and negative staining for thyroglobulin and PAX-8, which exclude thyroid origin and confirm lung origin. With regards to primary lung lesions, papillary adenoma is very rare with less than 30 cases reported [3]. It is worth mentioning that very few primary lung adenocarcinomas with morphological features of PTC have been reported [4, 5]. The present case was organ confined and showed no morphological features of malignancy. The proliferation rate, as assessed by ki67, was very low. Based on these characteristics, if the lesion was located in the lung, it would probably have been diagnosed as a papillary adenoma. However, when papillae show a complex architecture, the differential diagnosis with adenocarcinoma should be considered. Based on strict morphological criteria another differential diagnosis would be with glandular papilloma. This would be of academic interest because these lesions might belong to the spectrum of the same disease process. However, the morphological criteria used in lung pathology cannot be applied with confidence in the case reported herein. In view of the clinical context, a precise name cannot be assigned to this entity and the most appropriate term would be pulmonary-derived papillary adenomatoid proliferation.

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Conflict of Interest

The authors declare no conflict of interest.

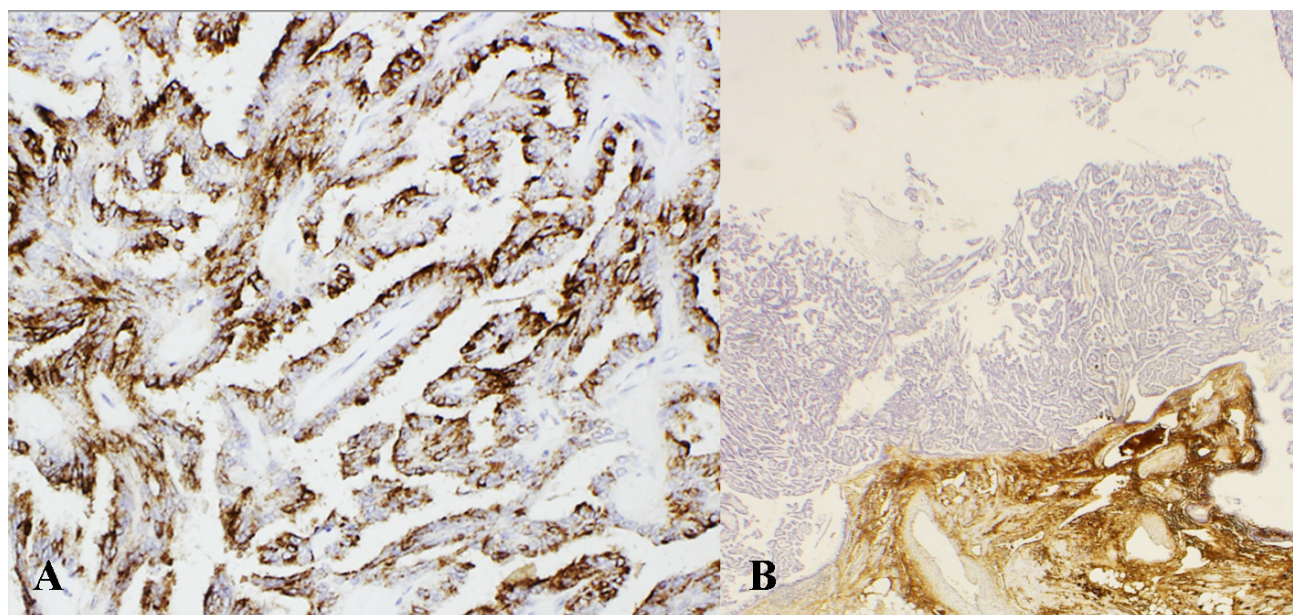


Figure 3. — Immunohistochemistry shows positive staining with surfactant apoprotein A (A) and negative staining with thyroglobulin with positive internal control (B).

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