

Adrenal heterotopic tissue in the peritoneum coincident with serous surface carcinoma

Ayşe Filiz Avsar,¹ Gülin Feykan Yeğın Akçay,² Emre Erdem Tas,¹ Huseyin Levent Keskin,² Aylin Yazgan³

¹Department of Gynecology and Obstetrics, Yıldırım Beyazıt University, Ankara; ²Department of Gynecology and Obstetrics, Atatürk Education and Research Hospital, Ankara; ³Department of Pathology, Atatürk Education and Research Hospital, Ankara, Turkey

Abstract

We report a case involving the concomitant presentation of two rare conditions: heterotopic adrenal tissue in the pelvic peritoneum and serous surface papillary carcinoma. A postmenopausal woman with an abdominal cystic mass and generalized ascites underwent laparotomy with the suspicion of ovarian carcinoma. Final histopathological analyses revealed serous surface carcinoma with metastasis to the endocervical canal, bilateral fallopian tubes, omentum, and para-aortic lymph nodes. Heterotopic adrenal tissue was also detected in the peritoneum.

Serous surface carcinoma of the peritoneum should be considered in the differential diagnosis when ascites, omental caking, and peritoneal nodules are observed in a patient with or without an ovarian mass. Heterotopic adrenal tissue is another rare condition in adults and can be detected incidentally in the peritoneum.

Introduction

Adrenal rest is common in children, especially in neonates, but it occurs in only 1% of adults.¹ It is usually identified incidentally during surgical procedures related to other conditions. Reported localizations of ectopic adrenal tissue include renal parenchyma; bladder; broad ligament and ovary; pulmonary, hepatic, and intracranial lesions; testis; spermatic cord; and celiac axis.^{2,3} We report a case of heterotopic adrenal tissue in the pelvic peritoneum coincident with serous surface papillary carcinoma, a rare tumor.

Case Report

A 63-year-old postmenopausal woman presented to our institution with abdominal distension and pelvic pain. She had a history of hypertension and hypothyroidism. Her family history was noncontributory. Her blood pressure was regular, with amlodipine.

Physical examination revealed a tender right abdominal mass and generalized abdominal distension caused by ascites. Routine blood indicators, liver and renal function, and fasting blood glucose level were in normal ranges. The patients' thyroid-stimulating hormone (TSH) level was 50 μ U/mL because of uncontrolled medications, and her cancer antigen 125 level was 55 U/mL. The initial diagnosis was a pelvic malignant mass of unknown origin.

On ultrasonography showed an irregular, multilocular cystic abdominal mass and generalized abdominal ascites. Computed tomography of the abdomen showed a 159×128-mm cystic mass with papillary projections and solid component, originating in the right adnexal area. Multiple peritoneal implants were also observed; the largest measured 30 mm and was located in the right lower quadrant. Based on these findings, the radiological diagnosis was ovarian carcinoma. To relieve abdominal pressure and obtain an ascites sample for diagnostic analysis, paracentesis was performed. The procedure reduced the patient's complaints, but no malignant cell was detected in the ascites sample.

After regulation of TSH, the patient underwent laparotomy with a suspicion of ovarian carcinoma. Findings from a frozen section of the right adnexal mass indicated high-grade serous carcinoma. Surgery comprising total abdominal hysterectomy, left salpingo-oophorectomy, bilateral pelvic para-aortic lymphadenectomy, appendectomy, and omentectomy was performed. Final histopathological analyses revealed serous surface carcinoma with metastasis to the endocervical canal, bilateral fallopian tubes, omentum, and para-aortic lymph nodes without ovarian involvement. Heterotopic adrenal tissue was also detected in the peritoneum (Figure 1).

Correspondence: Gülin Feykan Yeğın Akçay, Ufuk Üniversitesi Cad. No: 30/30 Cukurambar, Ankara, Turkey, Postal Code: 06610.
Tel.: +90.312.291.25.25/3839 - Fax: +90.312.291.27.26.
E-mail: gulinyegin@hotmail.com

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Discussion

Accessory adrenal tissue is found in 50% of post-mortem examinations in neonates and children, but as maturity leads to atrophy, rare in adults.¹ The adrenal gland has a dual embryological origin: the cortex arises from the coelomic mesothelium and the medulla arises from neural crest ectoderm. During migration of the medulla to the cortex, fragments of tissue can be separated, forming accessory adrenal glands. Heterotopic adrenal tissue can be found in close relationship to the sex organs because of the spatial relationship between the adrenal primordium and genital ridge in early embryogenesis.^{1,2}

The adrenal rest may be detected anywhere along the path of embryonic migration, including the celiac axis, genitals, and broad ligaments.⁴ The most common site of adrenal tissue is the spermatic cord region,⁵ but the incidence of this manifestation in adults is only 1%.⁶ Based on the migration theory, adrenal tissue may also be found in the genital system. In 2014, a case of oncocytic adrenocortical neoplasm arising in adrenal rest of the broad ligament of a 29-year-old Japanese woman was reported.⁷ Another rare case was a non-secreting pheochromocytoma of the broad ligament revealed by appendicular peritonitis.⁸

The clinical significance of heterotopic adrenal tissue is usually minimal, and this tissue is not typically associated with endocrine abnormalities.² Compensatory functional hypertrophy may also be seen in patients who have undergone bilateral adrenalectomy.⁹ Macroscopically, adrenal rests appears as a round, yellow nodule with a firm consistency, resembling a fat lobule.¹⁰ Malignant transformation is extremely rare, and only several cases (involving the renal hilus, spinal region, and testis) have been reported.⁴

The coincidence of genitourinary tumors and adrenal rest is rare. Only four cases of such tissue concomitant with testicular malignancies and an ovarian serous cystadenoma have been reported.^{2,5,11,12}

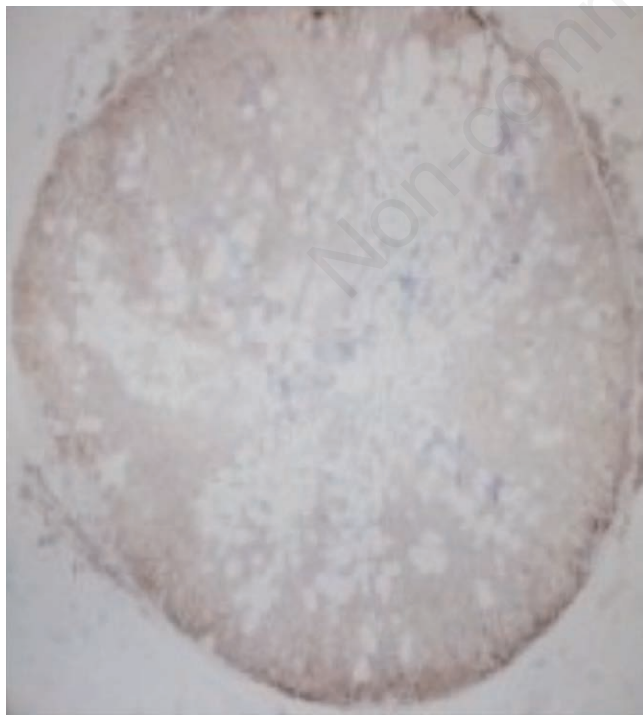


Figure 1. Heterotopic adrenal tissue contains myelipoma focus showing positive staining with Melan_A in peritoneal adipose tissue (magnification: x40).

Here, we report a case of heterotopic adrenal tissue localized in the pelvic peritoneum and coincident with serous surface papillary carcinoma. Serous surface carcinoma of the peritoneum is a rare, aggressive malignancy defined as a primary tumor that is histologically indistinguishable from serous carcinoma of the ovary and diffusely involves the peritoneal surface but spares or only superficially invades the ovaries.¹³

This case was diagnosed incidentally during histological examination of the surgical specimen. As the patient had no history of adrenal surgery, the presence of heterotopic tissue was due to the malpositioning or self-differentiation of cells during the embryonic period. The novelty and clinical significance of this case are related to the localization of adrenal rest and coincidence with serous surface carcinoma.

Conclusions

The case presented here is a report of the concomitant occurrence of two rare conditions: heterotopic adrenal tissue and serous surface papillary carcinoma. Serous surface carcinoma of the peritoneum should be considered in the differential diagnosis when ascites, omental caking, and peritoneal nodules are observed in a patient with or without an ovarian mass. Heterotopic adrenal tissue is another rare condition in adults and can be detected incidentally in the peritoneum.

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