Adrenal heterotopia in the fallopian tube: A case report
Heterotopia da suprarrenal na trompa de falópio: Um caso clínico

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Abstract

A case of an adrenal heterotopia in the fallopian tube is presented. It's an uncommon benign pathology found in a 73-year-old patient, accidentally identified in the pathologic specimen of a Wertheim-Meigs hysterectomy performed as treatment of endometrioid adenocarcinoma of the endocervix. When the patient was 47 years old, she was diagnosed with breast cancer and underwent a mastectomy and 3 sessions of chemotherapy. To our knowledge, this is the third case described in English literature reporting an adrenal heterotopia in the fallopian tube, and the first one associated with endometrioid endocervical adenocarcinoma and a history of breast cancer.

Keywords: Heterotopia; Ectopia; Adrenal; Gynecology; Fallopian tube.

Resumo

Neste artigo é apresentado um caso de uma heterotopia da suprarrenal na trompa de falópio. Esta é uma patologia benigna e rara, encontrada numa doente de 73 anos, acidentalmente identificada na peça operatória de uma histerectomia de Wertheim-Meigs, realizada como tratamento de um adenocarcinoma endometrióide do endocolo. Aos 47 anos, a doente foi diagnosticada com cancro da mama e foi submetida a uma mastectomia e 3 sessões de quimioterapia. Até onde sabemos, pela pesquisa de artigos escritos em inglês, este é o terceiro caso que relata uma heterotopia da suprarrenal na trompa de Falópio, e o primeiro associado a um adenocarcinoma endometrioide do endocolo e a história de cancro da mama.

Palavras-chave: Heterotopia; Ectopia; Suprarrenal; Ginecologia; Trompa de Falópio.

INTRODUCTION

The first description of adrenal heterotopia dates to 1740, when Morgagni, during an autopsy on a child, described the presence of yellowish nodules, similar to adrenal tissue, on the spermatic cord[1]. Later, many cases were reported, involving several different locations, mainly in the abdominal-pelvic area, more often in areas related to the kidneys and urogenital structures[2-5]. Other locations where there have been reported cases are: liver, lungs, pericardium, intracranial, intraspinal, gastric wall, pancreas, colon, celiac plexus, testicles, broad ligament, placenta, ovaries, and retroperitoneal zone[4,6,7].

Adrenal heterotopy has been reported in up to 50% of autopsies performed on newborns, but in more recent studies it was found that the incidence ranges from

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1% to 9.3% in pediatric patients, who have been surgically intervened, with cryptorchid, communicating hydroceles, and inguinal hernias, with a subsequent exploration of the area. In adults, it is estimated that the incidence is only 1% and may be the result of involution, superficial dissections, differences in diagnosis, or simply because it's harder to identify in the pelvic area of the adult[8,9]. It is usually found through autopsies or surgeries performed in the groin area. The incidence in female adults is even lower and is most often found accidentally after hysterectomies[8,10,11].

This article describes a rare case of an Adrenal Heterotopy, located in the left fallopian tube, which was found after a hysterectomy, performed to treat an endometrioid adenocarcinoma of the endocervix, a situation already considered rare.

CASE DESCRIPTION

A 73-year-old Caucasian female patient came to an appointment by postmenopausal metrorrhagia and pain in the hypogastrium. She has a personal history of breast cancer and underwent a mastectomy at the age of 47, on the left breast, followed by 3 adjuvant chemotherapy sessions. It wasn't possible to identify the breast cancer histological type and the adjuvant therapy drugs. As other medical antecedents, she reported hypothyroidism, gastritis, osteoporosis, and arterial hypertension. The medication taken by the patient was: Levothyroxine, Bioflavonoids, Irbesartan, Furosemide, Iron, and Vitamin D.

Upon gynecological examination, a friable endocervical polyp was detected and biopsied. The histological and immunohistochemical exam was consistent with the diagnosis of endometrioid adenocarcinoma.

For staging, a Magnetic resonance imaging was performed, in which a neo formative lesion of the cervix, centered on the endocervix, was reported, measuring 20 mm in the longest dimension. In the middle region of the cervix (also corresponding to the middle region of the tumoral lesion), disruption of the stromal ring of the cervix was defined, in a pattern suggesting local and parametrial invasion. No expansive lesions in the endometrium and adnexal regions were reported and no inguinal or iliac adenomegalies, ascites, or pelvic hydro-ureters were described. The tumor was then classified as endometrioid adenocarcinoma of the endocervix, in stage IIB of FIGO. As a treatment, primary chemoradiotherapy followed by Wertheim-Meigs hysterectomy with pelvic and lumbar-aortic lymphadenectomy was planned.

The study of the surgical specimen showed residual endometrioid adenocarcinoma at the cervix with no lymph nodes metastasis, no parametrial infiltration, and endometrioid intraepithelial neoplasia of the endometrium. In the left fallopian tube, a 2mm adrenal heterotopy was identified, composed of cells morphologically resembling the three layers of the adrenal cortex (glomerulosa, fasciculata, reticular) and which had positive immunoexpression to AE1/AE3 (focal) and inhibin (Figure 1).

At follow-up, 1 year and 5 months later, the patient didn't show any clinical signs of suspected local recurrence or distant spread.

DISCUSSION

We describe a clinical case of adrenal heterotopy accidentally diagnosed in a woman, at an advanced age, in a specimen of radical hysterectomy for endometrioid adenocarcinoma of the endocervix. Until 2018, in the literature, there were no reported cases of adrenal heterotopy in the fallopian tubes. A second case was published in 2021 combined with salpingitis isthmica nodosa[12]. Another particularity of this case is the fact that our patient had a background of breast cancer treated by mastectomy and adjuvant chemotherapy and also recent chemoradiotherapy for treatment of endometrioid adenocarcinoma of the endocervix. Both these exposures didn't destroy that adrenal heterotopy, suggesting the benignity of the lesion.

Some authors believe that adrenal heterotopias may originate in cells with the potential of adrenal tissue differentiation. However, the most frequently described explanation for heterotopia is that, during embryologic development, there is an invasion of elements of adrenal medulla through the cortex, originating fragments of adrenal tissue, that later migrate following genitals development[3,13].

The adrenal heterotopy commonly consists of a thin yellowish nodule of about 1 to 5 mm in diameter. Of the three adrenal gland cortical layers, fasciculate and

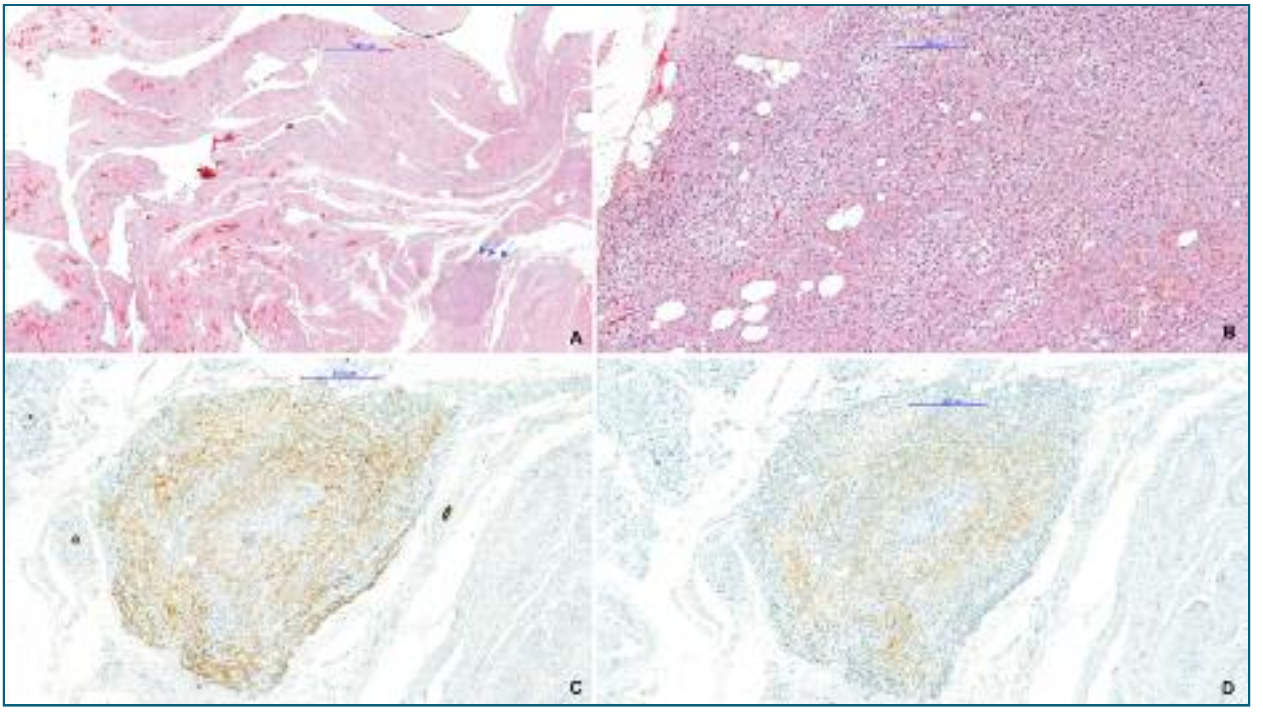

FIGURE 1. Heterotopic adrenal cortical tissue. (**A**) Microscopic examination revealed a small nodular lesion in the dependence of the fallopian tube (arrows), HE. (**B**) The lesion was composed by cells that resemble the three layers of the adrenal cortex, HE. The immunohistochemical study showed: (**C**) positivity for AE1/AE3 and (**D**) for inhibin.

glomerulosa predominate[14]. Usually, with age, due to the normal function of the adrenal gland, the heterotopic tissue will atrophy, as it does not need to produce hormones. Generally, when they persist until adulthood, they don't cause symptoms, but can increase in size and become symptomatic, releasing hormones that cause morbidity and mortality in affected patients[15,16]. Several clinical consequences of functionally active adrenal heterotopia have been described, including hyperplasia of the heterotopic tissue after primary adrenalectomy; adrenal insufficiency if the heterotopic tissue is inadvertently removed during a nephrectomy or resection of another organ; and benign or malignant neoplastic transformation[1,7].

In some cases, the heterotopic tissue can be hyperfunctioning and cause excessive hormonal production that leads to a set of clinical manifestations dependent on the type of hormone produced in excess. In addition to a hyperfunctioning state, heterotopic tissue can rarely undergo malignant transformation. Cases of pheochromocytoma, Leydig cell tumor and adenocarcinomas have been reported[17,18].

Being mostly of little size, non-functional and causing no symptoms, makes adrenal heterotopy tissue difficult to identify by clinical, hormonal, or imaging studies. When suspected, diagnostic imaging exams can help to locate adrenal heterotopies and help differentiate between benign or malignant tumors. For this purpose, magnetic resonance imaging seems to be the best imaging technique[19].

Usually, adrenal heterotopy is found accidentally in surgeries performed for other reasons. If clinically detected, resection is recommended because of the risk that adrenal heterotopia will turn into hyperplasia or neoplasia[8]. Usually, complications do not occur because it's easy to remove[20].

CONCLUSION

The case described in this article of an adrenal heterotopy is considered rare since it's found in a woman, of advanced age (73 years), and the heterotopy is found in the fallopian tube, associated with endometrioid

adenocarcinoma of the endocervix, findings that were never described in the literature.

As expected, the adrenal heterotopia proved resistant to the adjuvant chemotherapy for breast cancer treatment and to the chemoradiotherapy of endometrioid adenocarcinoma of the endocervix treatment.

As a result of the rarity of adrenal heterotopy, especially in women and in the absence of malignancy and hormonal functionality of the disease, prognostic implications are poorly understood. In short term, the risk of recurrence seems to be low, so imaging follow-up may be dispensed. There is scarce scientific information about health long-term implications, especially with the risk of future degenerative diseases such as cardiovascular, cerebrovascular disease or dementia, so a long-term follow-up of these patients seems justified.

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CONSENT

Written informed consent was obtained from the patient for publication of this case report.

CONFLICT OF INTEREST

The authors declare that they have no conflict of interest.

CONFIDENTIALITY OF DATA

The authors declare that they have followed the protocols of their work center on the publication of data from patients.

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