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Case Report

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Male Structures in Cystic Teratoma of the Ovary

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Abstract

Structures arising from the male urogenital sinus are rarely observed in ovarian mature teratomas. We refer a case of a 31-year-old woman surgically treated for a monoliteral ovarian cyst. Histological examination revealed an ovarian teratoma with a focus of prostate gland. The nature of the cells was confirmed by the positivity for PSA, PAP and CK34BE12 antibodies. The specific cells expressed receptors for Estrogens and Progesterone. The presence of prostate tissue in a mature cystic teratoma of the ovary has been correlated with genetic and/or embryological theories.

Keywords: Cystic Teratoma; Male Structure; Ovary; Prostate

Introduction

Mature cystic teratoma is a benign germ cell tumor more frequent among women of childbearing age. It is composed of a variety of mature tissues derived from the three germ layers (Endoderm, Mesoderm and Ectoderm). Some tissues (skin and dermal appendages, bone, fat, intestinal, gastric and respiratory epithelium, cartilage, muscle and nerve tissue) occur more frequently than others (thymus, salivary glands, kidney and pituitary gland). In teratomas, structures arising from the male urogenital sinus are occasional findings, there being only 28 published cases with prostate tissue.

Case Report

We report a case of a nulliparous woman of 31 years of age, who underwent surgery for suspected ovarian neoplasm.

Methods

The sample was fixed in 10% neutral buffered formalin, and representative samples were embedded in paraffin and stained with hematoxylin-eosin and with Mallory trichrome.

Immunohistochemically analysis was performed on paraffin inclusions using the following antibodies:

- PSA (monoclonal antibody, 1:200, Novocastra)
- PAP (polyclonal antibody, 1:100, Neomarkers)
- CK 34βE12 (monoclonal antibody, 1:100, Dako)
- ER (monoclonal antibody, 1:50, Novocastra)
- PR (monoclonal antibody, 1:40, Novocastra)

Lesion Examination

Macroscopically, the lesion was 6 centimeters in diameter and was composed of two contiguous cysts, one with a thick wall containing pilo-sebaceous material and one containing mucoid substance. Histologically, the lesion was composed of solid and cystic areas, containing pilo-sebaceous material, lined by an epithelium of epidermal and respiratory type, with relative skin appendages. There was also residual ovarian tissue including a minute mucinous cystadenoma.

A small not encapsulated nodular area, in the solid component of the tumor, showed glandular acini lined by a single layer of cubic or cylindrical epithelium, with clear or scant cytoplasm and with small, round, basal or central nuclei, without evident nucleoli. There were also rare cells of the basal layer, sometimes hyperplastic. (Figure 1) These glandular acini were organized in lobules separated by a fibromuscular stroma

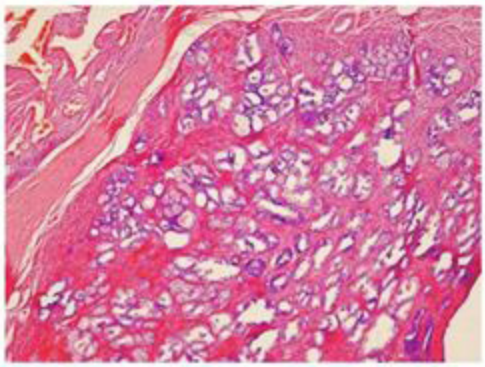

Figure 1: Prostatic glands in ovarian teratoma. The lobules are intersected by a different amount of smooth muscle cells. The epithelium shows in several unities an enhancement of basal cells.

80% of the luminal glandular cells, immunostaining was positive for PSA and PAP, with a uniform and regular pattern, and CK34βE12 stained the rare cells of the basal layer (Figures 2, 3).

The novelty found in our case concerns the positive estrogen receptor and progestin only in the structures related to the prostate elements than to the other tissue Microscopic aspects and immunophenotyped confirm that it was prostate tissue. The pattern of immunopositivity for PSA and PAP also allows the prostate gland to be distinguished from female paraurethral glands (Skene's Glands), in which these two antibodies have a multifocal and irregular staining [1].

In our case, differently from other cases reported in literature [2,3] we did not find either tissues derived from male urogenital sinus or cells producing androgens (such as luteinized type cells, etc.).

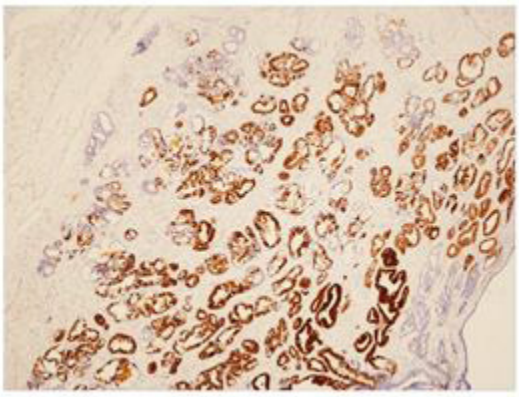

Figure 2: Immunohistochemically positivity of the glands for the PSA Antigen.

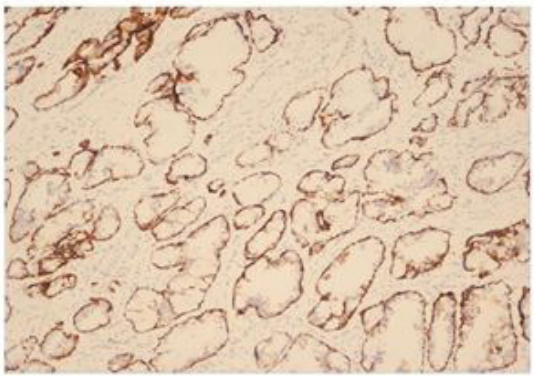

Figure 3: The cytokeratin 34-beta E-12 is expressed by the basal cells.

Discussion

Despite the wide variety of tissues found in mature cystic teratomas, the presence of prostate tissue is occasional and probably underestimated [4], with only 28 cases reported in the literature [5-10]. These are generally women of childbearing age (average 35 years) with ovarian masses of average diameter of 7.3 cm and weight of 153 grams, located mostly in the left ovary. Areas of prostate tissue have an average diameter of 1.2 cm, and in some cases invasive adenocarcinoma [11] or adenocarcinoma *in situ* [12] were found.

The presence of masculine tissues in ovarian teratomas is not yet a fully understood phenomenon: these tumors arise from a single ovarian germ cell by parthenogenesis, after the first meiotic division [1,2,13] and these cells generally have a 46 XX karyotype, or sometimes a 46 XXX trisomy [11].

Embryologically, the prostate gland (as other glands and structures of the male genitals) comes from the male urogenital sinus, derived from the fusion of the euro-rectal septum with the terminal mesospheric ducts close to the origin of the ureters. From this structure, at 30-35 days of gestational age, the urethra, the prostate and the lower part of the vesicle are derived. The Y chromosome is necessary for male sexual differentiation, its absence therefore excludes the development of such tissues in ovarian teratomas; for this reason, three pathogenetic theories have been proposed [5]. According to the first hypothesis, prostate tissue may result from residual embryonic endodermal sinus stimulated by androgens, produced locally by luteinized cells near the teratoma [11,13,14]. Or else, these tissues could result from metaplasia of mesonephric remains in an androgenic microenvironment [11,15]. Or thirdly, the role of '"imprinting" of paternal X has been suggested, escaping from inactivation mechanisms [16].

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