Bilateral androblastoma (Sertoli-stromal cell tumor) of the ovary: a rare cause of virilization in a teenager

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Summary

A case of a 17-year-old patient diagnosed with bilateral androblastoma of the ovary is presented. The patient was admitted because of secondary amenorrhea, hirsutism and acne. After clinical, ultrasonographic and hormonal examinations an androgen-producing ovarian tumor was suspected and consequently laparotomy with right ovarian tumor excision and left ovary exploration was carried out. During surgery the right ovarian tumor was excised and exploration of the left ovary revealed an ovarian tumor with a diameter of 10 mm, which was then also excised. The pathologic diagnosis was a bilateral androblastoma of the ovary measuring 40 mm x 30 mm x 20 mm in the right ovary and 10 mm in diameter in the left ovary. We concluded that androblastomas, in spite of their low incidence, are a possibility that should always be considered in women of all ages presenting with signs of virilization.

Key words: Androblastoma; Virilization; Teenager.

Introduction

Androblastomas (Sertoli-stromal cell tumors) are a rare cause of female virilization. The tumors are known to contain Sertoli cells, Leydig cells, indifferent stromal cells or all three cell types in various proportions and varying degrees of differentiation [1]. They occur at all ages but are most commonly encountered in women during their early reproductive years [2]. They are usually unilateral, and bilateral androblastomas are present in only less than two percent of cases [3].

Case Report

A 17-year-old patient with an unremarkable medical history was admitted to our department because of secondary amenorrhea, hirsutism and acne. Menarche was at the age of 13, however menses were irregular (occurring every 2-6 months) with the last menstruation present at the age of 14. Physical examination revealed a normal body mass index, an increase in hair growth on the abdomen, thorax, upper limb and chin (17 points on the Ferriman-Gallwey scale), acne on the face and thorax, voice deepening and clitoromegaly. Transvaginal ultrasound (TVS) showed a normal uterus, normal endometrium (4 mm), a right ovary measuring 44 mm x 30 mm with a well defined hyperechoic lesion measuring 30 mm in diameter, a left ovary measuring 38 mm x 20 mm with a hyperechoic internal part and 12 small peripheral follicles (8-10 mm in diameter). Analytical tests revealed a high testosterone serum level of 2.96 ng/ml [0.06-0.82] and a low FSH of 2.31 IU/l [3.5-12.5]. All other hormonal tests (LH, 17β-estradiol, prolactin, sex hormone binding globulin, dehydroepiandrosterone sulfate, TSH and thyroxin) were within normal ranges. After clinical and ultrasonographic examinations an androgen-producing ovarian tumor was suspected and consequently laparotomy with right ovarian tumor excision and left ovary exploration was carried out.

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During surgery the right ovarian tumor was excised and exploration of the left ovary revealed a left ovarian tumor with a diameter of 10 mm, which was then also excised.

The pathologic diagnosis was a bilateral androblastoma of the ovary measuring 40 mm x 30 mm x 20 mm in the right ovary and 10 mm in diameter in the left ovary.

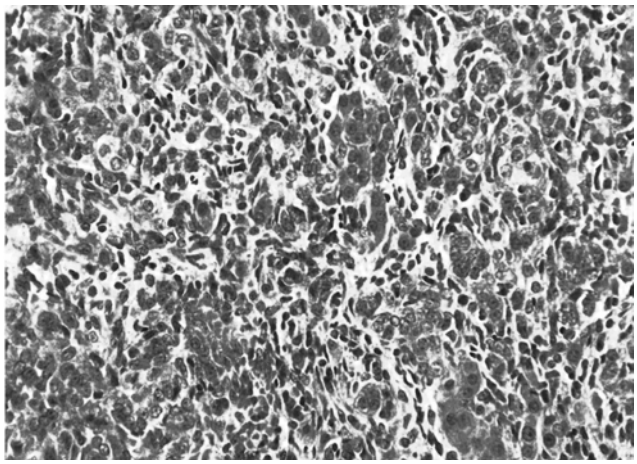
Both tumors were localized in the cortical part of the ovaries. The larger one was partially covered by an ovarian capsule and loose connective tissue. Sectioning revealed a solid tumor with a brown-yellow appearance. Upon microscopic examination Sertoli cells were observed forming solid tubular structures with focal hollow tubules and solid islands. Leydig cells were also found in abundant amounts - forming distinct areas of focal groups inbetween the Sertoli cells. The tumors were both well differentiated without mitotic activity, therefore classifying them both as Stage IA (Figures 1 and 2).

Postoperatively the patient presented with no complications. Two days after the operation the level of testosterone was 0.21 ng/ml [0,06-0,82]. The patient was then followed up within one month, at which time she reported the occurrence of spontaneous menstruation and a discreet improvement in her hirsutism and acne severity. In the analytical control the level of testosterone was 0.36 ng/ml [0.06-0.82]. TVS showed no abnormalities with regard to the uterus and ovaries.

Discussion

In this case the main clinical features and reasons for consultation of the patient were secondary amenorrhea and symptoms of severe hyperandrogenism. Secondary amenorrhea, when it presents in young women, may be due to variety of causes such as hypothalamic insufficiency, hyperprolactinemia, hyperandrogenic syndromes, and may also be due to primary ovarian insufficiency. Hyperandrogenic symptoms may have an ovarian or suprarenal origin. Among ovarian causes polycystic ovarian syndrome is the predominant one. Tumors of the ovary associated with hyperandrogenism include primitive tumors of the sexual cords and stroma (granulosa cell

Fig. 1



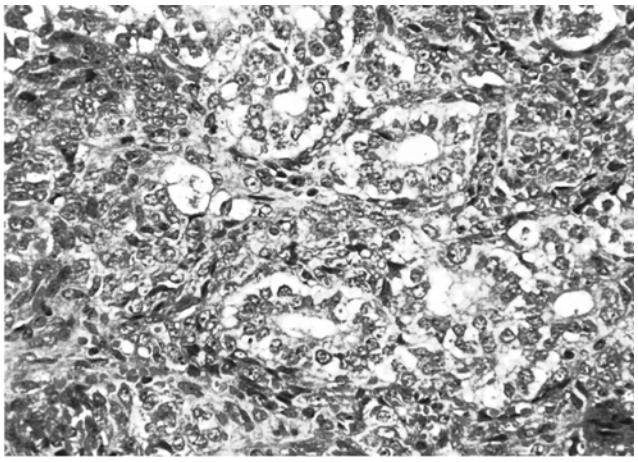


Fig. 2

Figure 1. — Leydig cells with abundant eosinophilic cytoplasm and Sertoli cells dispersed among them.
Figure 2. — Hollow tubules with Sertoli cells.

tumors, thecomas, androblastomas), steroid cell tumors (luteomas and Leydig tumors), and primary and secondary ovarian tumors which contain functioning stroma [4].

Androblastomas are rare ovarian tumors; they can occur at all ages but are most often encountered in women in their early reproductive period with an average age of diagnosis found at 25 years of age [5]. Virilization occurs in more than one-third of cases and plasma testosterone levels are always elevated in the virilized patients. The levels generally range from 1.2 to 7.0 ng/ml [5], and weaker androgens as well as other steroid hormones may also be increased to varying extents [5, 6]; 17-KS values are usually normal or only slightly elevated.

The diagnosis of androgen-producing ovarian tumors is made using clinical, biochemical and imaging techniques. However, the imaging techniques are often insufficient because the tumors are usually very small. Therefore, if an ovarian tumor is suspected clinically and biochemically, a diagnostic and surgical laparoscopy is indicated and should be performed.

After the removal of a virilizing androblasoma normal menses usually resume in about four weeks. In most cases the hirsutism improves but often incompletely. However, clitoromegaly and particularly deepening of the voice are less likely to regress.

In conclusion, androblastomas of the ovary are rare, most are benign, and they do require surgical treatment. However in spite of their low incidence, they are a possibility that should always be considered in women of all ages presenting with signs of virilization.

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