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Premature Puberty Revealing an Ovarian Tumor in a Five-Year-Old Girl

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Abstract

Background: Ovarian tumors in the girl child are sometimes revealed by the development of secondary sexual characteristics. The authors report the case of a five-year-old girl in whom the disease was revealed by early puberty. **Case presentation:** A five-year-old girl with an enlarged abdomen for about four months. The onset of pain and the sensation of a mass prompted the consultation. The development of secondary sexual characteristics (SSC) noted by the family had not been mentioned. The patient was classified as pubertal stage 2 according to the Tanner classification. An abdominal ultrasound and a CT scan showed a large left ovarian mass, an enlarged uterus for the patient's age and a normal right ovary. The hormonal workup was not contributive. The treatment consisted only of a left salpingo-ovarectomy, without complementary chemotherapy. Anatomic pathological examination of the surgical specimen concluded to a juvenile tumor of the granulosa. The evolution was good with a beginning of regression of the HSC one month after the ovarectomy. **Discussion:** Granulosa tumors are sometimes secretory cancers, generally with a low potential for malignancy and therefore a very good prognosis. Surgery based on total adnexectomy is the first-line treatment. The large size of the tumor, the presence of ascites and capsular rupture are factors of poor prognosis, hence the importance of early diagnosis. **Conclusion:** Routine comprehensive physical examination should be de rigueur for abdominal masses in girls, especially in the context of various beliefs that may impede early referral to care.

Keywords

Ovarian Tumor, Juvenile Granulosa Tumor, Early Puberty, Early Diagnosis,

Surgery

1. Introduction

Malignant ovarian tumors in young girls are rare and dominated by malignant germ cell tumors [1] [2]. Juvenile granulosa tumors derive from sex cord cells and are sometimes revealed by early pseudopuberty, which is usually isosexual. The early development of secondary sexual characteristics, associated with the increase in the volume of the abdomen, allows the diagnosis to be evoked. These clinical arguments are important in the context of limited resources, where the availability and accessibility of hormone assays are not always effective. The authors report the case of a five-year-old girl in whom the diagnosis of an ovarian tumor was suspected in view of the occurrence of precocious pseudopuberty.

2. Case Presentation

Five-year-old girl, the last of three children, with no particular personal or family history. The patient had been referred for an abdominal mass noticed by parents about two months before. The history of the disease was marked at the beginning by intermittent abdominal pain of the compression type, without intestinal transit disorders or vomiting; a secondary increase in the volume of the abdomen would have motivated a consultation and referral to the hospital.

On admission, the general condition was preserved; the skin and mucous membrane staining was normal and the nutritional status was good. Palpation of the abdomen revealed an ovoid, firm, poly-lobed mass extending from the right flank, protruding five cm beyond the umbilical region on the left, with a long axis of about 20 cm. There was no sign of ascites. The examination also showed development of the external genitalia, corresponding to Tanner stage II (**Figure 1**). Extensive questioning revealed that the breast enlargement had been noted before the increase of the abdominal volume, but had not prompted a medical consultation. Growth was normal with a BMI/age Z-score above the median.

Abdominal-pelvic ultrasound showed a large, hypoechoic, heterogeneous left ovarian tissue mass with fluid pockets, estimated at 206 × 145 mm, rising below the midline; the uterus was increased in size for age (64 × 18 × 28 mm). The liver, kidneys, spleen, pancreas, gallbladder and bladder were normal in appearance. There was no ascites. Abdominal computed tomography (CT) revealed a large, well-limited left intra-ovarian tissue formation with a dual multiclonal fluid and tissue component, enhanced after contrast injection and persisting at late time; this mass measured 162 × 153 × 100 mm. The uterus was homogeneous and increased in size (55 × 36 × 28) mm. The hormonal workup performed showed normal levels for age of HCG, alpha fetoprotein, LH and FSH, and estradiol. Inhibin could not be measured. Bone age was not assessed. The clinical (exhaustive clinical examination) and paraclinical (thoracic abdominal-pelvic imaging) ex-

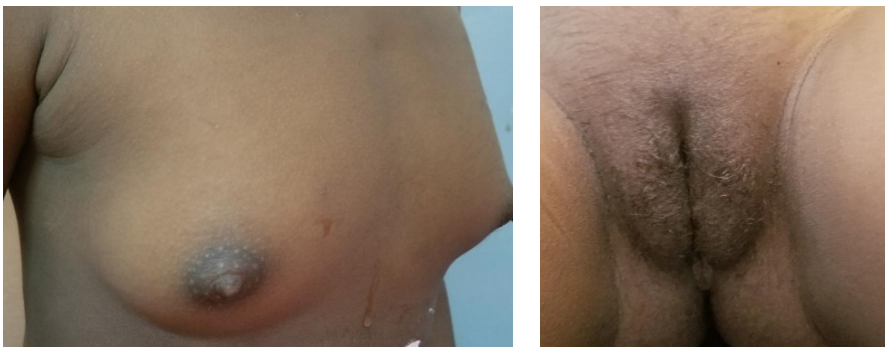
tension workup did not show any other tumor location.

The therapeutic management consisted of a left salpingo-ovarectomy, without neoadjuvant chemotherapy. The tumor was classified as stage Ia according to the FIGO clinical classification [3]. Pathological examination of the excisional specimen (**Figure 2**) concluded that it was a juvenile granulosa tumor.

The postoperative course was simple and the evolution was marked after two months by the beginning of regression of secondary sexual characteristics. Hormone assays, only available in private laboratories, were not repeated, due to the lack of financial means. There was no event after a current follow-up of 3 years.

3. Discussion

Ovarian tumors in children are rare, often organic, with a malignant contingent in 10% of cases. The malignant forms are sometimes secreting, leading to the early development of secondary sexual characteristics, depending on the type of hormones secreted. This is the case of juvenile granulosa tumors, originating from the sexual cords. They derive from gonadal interstitial tissue with endocrine function. They are rare types of ovarian tumors in young girls, in whom malignant germ cell tumors are more frequent [1]. Isolated cases or short series are often reported [4] [5] [6].



(a) (b)

Figure 1. Development of breasts (a) and hairiness (b) corresponding to TANNER stage II.

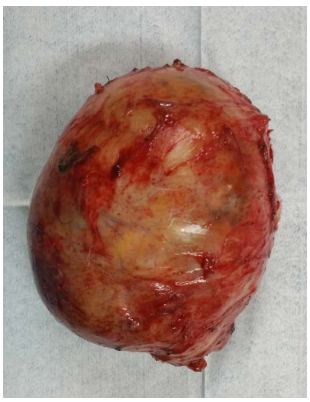


Figure 2. Photo of the surgical specimen (left adnexectomy).

Regardless of the histological type, ovarian tumors in young girls usually present with an abdominal symptomatology of pain, distension and enlargement, as in our patient. The enlargement of the abdomen is mainly related to tumor growth, but ascites of varying extent may also be present; it is taken into account in the FIGO clinical classification of ovarian tumors. Signs related to androgenic or estrogenic impregnation are only observed in case of secretory tumors; these tumors derive from the mesenchyme and the sexual cords. According to the WHO classification, these include granulosa and stroma tumors, and roblastomas, Sertoli and Leydig cell tumors, gynandroblastomas and other unclassified tumors [7]. Nevertheless, cases of juvenile granulosa tumors without signs of estrogenic or androgenic impregnation have been reported [5] [8]. Moreover, normal estradiol levels are sometimes observed even in the presence of precocious pseudopuberty, as in our patient. This could be explained by a circadian variation of estradiol secretion by the tumor cells. Moreover, a low specificity of plasma estradiol determination by conventional methods is sometimes evoked and could also contribute to this clinico-biological discordance [5].

Juvenile granulosa tumors generally have a good prognosis when diagnosed early [9]. In our patient classified as FIGO stage Ia, treatment consisted of left adnexectomy. Conservative surgery is sometimes discussed in early stages.

4. Conclusion

The case reported in this observation reminds us of the necessity of a systematic and exhaustive physical examination in the presence of any abdominal mass. The presence of secondary sexual characteristics in such a young girl quickly pointed to the ovarian location of the tumor and contributed to a relatively early diagnosis and therapeutic management of the patient. Progress remains to be made in the accessibility of hormonal assays, useful not only for diagnosis but also for post-therapeutic monitoring.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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