Excision of ectopic adrenocortical tissue during laparoscopy for isolated tubal torsion

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SUMMARY:
Excision of ectopic adrenocortical tissue during laparoscopy for isolated tubal torsion.


Ectopic adrenal tissue is an occasional finding during surgery procedures. These remnants are usually found near the primary gland or along the course of gonadal vessels consistently with embryologic development. The appearance is of a small but distinct, soft, round, yellow nodule and only cortical adrenal tissue is normally observed. We report a case of an emergency laparoscopic salpingostomy for isolated tubal torsion with excision of EAT, incidentally found in left ovarian pedicle, followed by postoperative resumption of regular menses and regression of hirsutism. Although intraoperative research of ectopic adrenal tissue is not necessary its removal is indicated in case of incidental finding. If hyperandrogenic features are present, ectopic adrenal tissue should be actively researched when surgery is performed.

KEY WORDS: Ectopic adrenal tissue - Laparoscopy - Tubal torsion.

Methods, techniques, drugs

Introduction

Ectopic adrenal tissue (EAT) is often observed among newborn infants and children despite being apparently rare in adulthood. Growing up leads to atrophy of EAT: while such tissue is found in 50% of post-mor-
common mesodermic origin with gonads could lead us to a deeper comprehension of EAT pathogenesis and recognition.

**Case report**

A 22-year-old nulliparous woman was admitted to our emergency room referring acute right-sided pelvic pain, nausea and vomiting. She was clinically stable and afebrile. Irregular menses were reported. Clinical examination revealed slight hirsutism but the patient was virgo and refused rectal examination. Ultrasound revealed an edematous, enlarged, tortuous, and cystic right fallopian tube and a moderate amount of free pelvic fluid. The uterus, left tube and both ovaries appeared normal. Differential diagnosis between pelvic inflammatory disease and torsion could not be made. Ultrasound revealed an edematous, enlarged, tortuous, and cystic right fallopian tube and a moderate amount of free pelvic fluid. The uterus, left tube and both ovaries appeared normal. Differential diagnosis between pelvic inflammatory disease and torsion could not be made. Magnetic resonance imaging revealed a tubular, tortuous, cystic right adnexal mass (6 x 3 x 4 cm) and normal right ovary. The mass was assumed as hydrosalpinx. Since pain was resistant to analgesics and a slight leukocytosis was evidenced at laboratory testing a diagnostic laparoscopy was performed in urgent setting (6, 7). We used Veress needle to induce pneumoperitoneum positioning a 10-mm trocars trans-umbilical for the camera and other three 5-mm trocars in left and right flank and in sovrapubic region (8). Laparoscopy demonstrated an hydrosalpinx with a 720° torsion of the right fallopian tube (Figure 1a). Drainage of the hydrosalpinx and salpingostomy was carried out. During the operation, a well-circumscribed nodule in the left infundibulopelvic ligament was found and excised. It was visible as a small yellow disk through the peritoneum (Figure 1b). She recovered uneventfully and was discharged on the third postoperative day. On gross pathology the cut-surface of the nodule appeared golden-yellow, hard and it measured 3 mm in diameter. Microscopically the same had a well defined thin capsule. All three layers of the adrenal cortex were identified, showing normal zonation (Figure 1c, 1d). Peritoneal cytology showed inflammation but no malignant cells. Postoperative endocrinological evaluations including an

![Figure 1 - a) Hydrosalpinx; b) ectopic adrenocortical nodule in the left infundibulopelvic ligament; c) the nodule had a well defined thin capsule; d) all three layers of the adrenal cortex were identified, showing normal zonation.](image-url)
adrenocorticotropin hormone stimulation test revealed no abnormalities in the adrenocortical and gonadal steroids secretions. Nonetheless, follow-up visit revealed a mild residual dilation of the right tube at pelvic ultrasound but regression of hirsutism and resumption of regular menses.

**Discussion**

Occasional finding of EAT in women along gonadal vessel may be explained by the strong embryologic link between adrenal cortex and gonads. The most widely accepted theory on the pathogenesis of EAT is the descent of fetal cortical cells together with gonadal tissues, in the 7th fetal week (9). The adrenocortical primordium is unencapsulated and develops close to the emerging gonad at its early stages. Therefore, it is not surprising that some cells of the adrenocortical primordium may become associated and migrate alongside the gonad, to be found postnatally distant from the adrenal in the path of gonad descent. In practice, accessory adrenocortical tissue is most often encountered around the adrenal glands themselves (10). It has also been reported in such several locations as the liver, kidney, pancreas, transverse colon, celiac plexus area, broad ligament, epididymis, rete testis, ovary, testicle and retroperitoneal along the course of the vessels to the gonads (11). EAT are generally constituted by adrenocortical cells. Medullary cells are seldom found in ectopic fragments and have been observed exclusively in proximity to the surrenal glands (12-14). The medulla has a neuroectodermal origin and represents only 10-20% of the mass of the adult adrenal gland. The independent neural origin of the adrenal medulla and its relatively late migration into the cortical mass by week 9 of gestation, may explain its absence from adrenal rests. Leibowitz supposes that, during migration of the medulla, fragments of tissue, most frequently cortex, can be separated, forming accessory adrenal glands. Some Authors believe that ectopic adrenal tissue may develop in situ from a totipotential cell (15). The ovarian pedicle and its insertion on the lateral pelvic wall are the most common location in women. Incidence in adult is variable in literature varying from 1% to 23.3%, on the basis of autopic examination and intraoperative findings (1, 11). This may be due to a different care in surgical dissection and pathology examination. In Fall series the median ectopic tissue diameter observed was 2 mm and this may explicate significantly higher incidence.

Like adrenal masses (16-18), EAT are usually asymptomatic, they can give clinical symptoms related to their secretory activity: high levels in aldosterone, cortisol and androgens may result in metabolic and electrolytic imbalance and virilization (14, 19-22). Rarely, EAT can show neoplastic transformation resulting in neuroblastoma, hemangioma, myelolipomas and oncocytomas (19, 23-28). Since the majority of EAT cases has no clinical relevance, their functional significance is not clear. Nonetheless, compensatory hypertrophy secondary to adrenalectomy have been reported (11).

To date there are no large series or detailed review on EAT occurring inside female pelvis and the majority of information can be retrieved by case reports. The gross appearance is of a small but distinct, soft, round, yellow retroperitoneal nodule, sometimes coated by fat tissue. Microscopically all cortical layers are present thus a well-developed zona fasciculata and zona reticularis are observed and the absence of any medullary tissue together with a central vein and a well-defined capsule are normally found. Our case in consistent with this pathological description. The postoperative regression of hirsutism and the resume of regular menses may related to EAT removal but no hyperandrogenism was demonstrated preoperatively.

**Conclusion**

Although intraoperative research of EAT is not necessary its removal is indicated in case of incidentally finding. Laparoscopy may help in the identification of such lesions and thus increase their incidence. If hyperandrogenic features are present, EAT should be actively researched when surgery is performed.

**References**

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