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Case Report

Autoamputation of the Left Ovary in a Child: A Case Report and Review of Literature

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Abstract

Ovarian auto-amputation is an extremely rare condition. Chronic adnexal torsion and subsequent devascularization which leads to infarction and necrosis is the most accepted theory to explain this clinical entity. Herein we present a female infant with an antenatally diagnosed abdominal mass which was found to be an auto-amputated ovary floating freely in the abdominal cavity after surgical intervention. The topic is also discussed under the light of relevant literature.

Keywords: *Ovarian Auto-Amputation; Children*

Introduction

Ovarian auto-amputation is a rare condition with an incidence of 1 in 11421 [1]. The first description of this entity was defined by Giovanni Morgagni in 1748 [2]. Chronic adnexal torsion and subsequent devascularization leading to infarction and necrosis is the most accepted theory to explain this rather rare clinical entity.

Here we present a female infant with an antenatally diagnosed mass in abdomen and was found to have an auto-amputated ovary floating freely in the abdominal cavity. The topic is also discussed under the light of relevant literature.

Case Report

A 4-month-old female infant was admitted to our clinic with an antenatally diagnosed abdominal mass. Prenatal ultrasonography (US) in the 32nd week of gestation revealed an abdominopelvic mass. She was otherwise normal. Abdominal examination revealed a mobile, nontender mass with a dimension of 5 x 6 cm. After her birth and before her admission to our department, she was followed with monthly sonographical examination for a possible involution or resolution of the mass. Former US before admission revealed that the mas was located in the left abdomino-pelvic region. Control sonography performed during her admission to our clinic revealed no resolution or involution in the mass but the mass was found to be located in the right abdominopelvic region revealing that the mass was freely mobile in the abdominal cavity. Postnatally T1 and T2 weighted magnetic resonance imaging (MRI) showed a hyperintense cystic lesion containing debris with a dimension of 54 x 34 x 55 mm (Figure 1). The laboratory tests including lactate dehydrogenase (LDH) and alpha fetoprotein (AFP) were within normal limits. Elective laparotomy under general anesthesia with a supraumbilical transverse incision revealed a free-floating, brown, round, smooth cystic lesion measuring 7 cm in diameter without an attachment to peritoneum or other abdominal viscera (Figure 2). The left ovary was absent and the left fallopian tube was found to be blind ended. Right ovary and right fallopian tube

was normal. Histopathologic examination showed that the excised specimen consisted of hemorrhagic, necrotic, autolytic tissue with no ovarian parenchyma. With an uneventful postoperative course, the patient was discharged from the hospital on the postoperative 3th day in good condition.

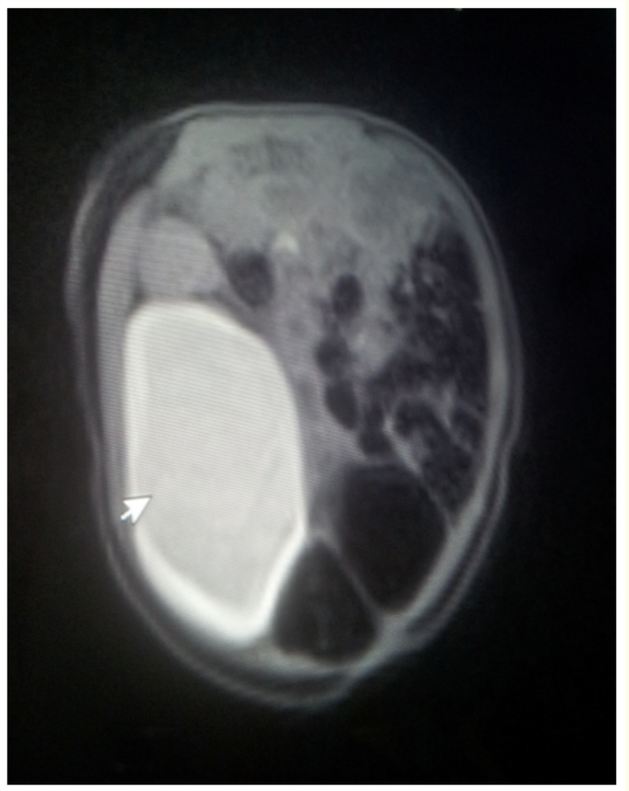

Figure 1: *Postnatal abdominopelvic MRI showing a hyperintense cystic lesion (Arrow: free-floating cystic mass).*

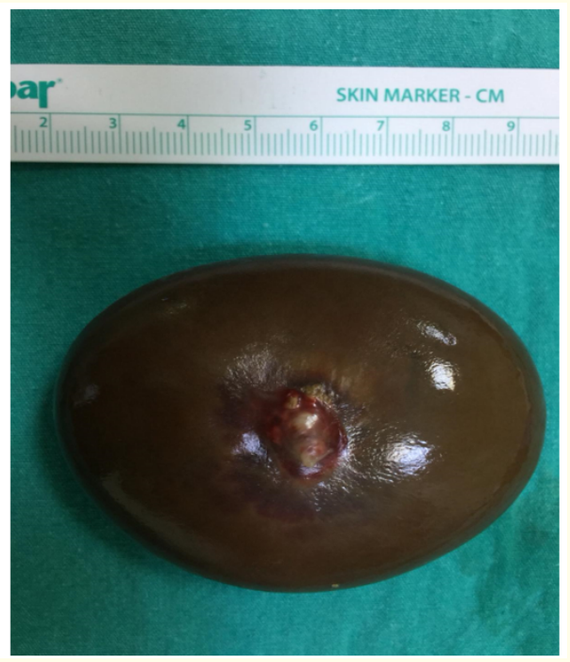

Figure 2: *Free-floating, autoamputated ovary and cystic mass.*

Discussion

The differential diagnosis of abdominal cystic masses in the perinatal period is broad and include intestinal duplication cysts, lymphangiomas, mesenteric cysts, cystic meconium peritonitis, neuroblastomas, and benign cystic teratomas, renal cystic dysplasia, urachal, mesenteric, omental and choledochal cysts, hydrometrocolpos, anterior meningocele and ovarian cysts. With a predominance in the female patients ovarian cysts constitute the most common pelvic/abdominal cystic masses [3-5].

A free-floating intraabdominal mass is an extremely rare condition and it almost always originates from ovary [6]. As an etiological factor regarding the involved organ apart from ovary, autoamputation of a gall-bladder and appendix epiploica following torsion was also reported but these patients are adults [7,8].

In 1748, after first description of an auto-amputated adnexa in a postmortem study by Morgagni, 50 more postmortem cases were reported by Ogorek in 1914 [9]. With the introduction and advancement of prenatal US, the first case of an antenatally diagnosed mass, after surgical intervention, which proved to be an auto-amputated adnexa was reported in 1983 [10]. To our best knowledge, there are approximately 28 cases reported in the literature who had cystic mass found during antenatal US and later on in postnatal life, at exploration, found to be auto-amputated ovary [11]. Our case is probably the 29th case reported in the English language literature.

As an entecedent event, many authors support the notion that, any type of vascular accident results in auto-amputated adnexa. Although some authors suggest the notion of a congenital absence of adnexa as an etiological factor, congenital absence is not accepted as an etiology for two reasons: 1) Mullerian structures arise from different embryological origins. 2) Absence of associated renal anomalies in reported series [2].

An auto-amputated ovary may be unilateral or bilateral. Right sided ovarian torsion is encountered more frequently [12]. Because of protective effect of sigmoid colon, left sided ovarian torsion is less common [13]. With a normal looking right fallopian tube and ovary with blind ended left tuba uterine and absent left ovary, our case, which was later on found to be a free-floating left torsed ovary, is contrary to the common notion that most ovarian torsion occurs in the right and our case also adds to the literature that left ovarian involvement may also be observed with respect to auto-amputated adnexa.

Another important point to mention in these patients is the necessity of exploration of contralateral adnexa. It is suggested that during a surgical intervention on tuba uterine and ovary, contralateral evaluation of the adnexa is mandatory if it is normal or present. If one takes the parental anxiety on the future fertility of their offspring in to account, at surgical intervention, if contralateral ovary and fallopian tube is found to be normal, relaxing explanations to the parents on the future reproductive capacity of their children is suggested and is useful in decreasing their anxiety [2].

It is important to distinguish between the diagnosis of adnexal torsion and auto-amputation. The diagnostic hallmark of an auto-amputated cyst or ovary is the detection of a freely mobile mass in the abdominal cavity as in our case [14]. Auto-amputation is irreversible. Adnexal torsion, on the other hand, is a medical emergency and once this diagnosis is considered, prompt surgical intervention becomes a matter of necessity rather than of choice for the protection of future reproductive capacity of these children [15]. During surgical intervention, it must be aimed to preserve compromised fallopian tube and ovary before torsion leads to hemorrhage and necrosis or auto-amputation.

Conclusion

In conclusion, auto-amputated adnexa is a rare finding in children. Many of these auto-amputations are asymptomatic and usually identified incidentally and do not compromise fertility if the contralateral adnexa are normal. It is important for the health providers to be aware that auto-amputation of the adnexa may be the end result of the fetal ovarian cysts diagnosed on antenatal US and should be kept in mind in the differential diagnosis of children with abdominopelvic cystic masses and be managed accordingly.

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