Case Report

Auto-amputated Ovarian Cysts in Infants : Current Status

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Auto-amputated Ovarian Cyst (AOC) is a rare finding in the pediatric population especially in infants, but it must be considered for the incidental finding of absence of the fallopian tube or ovary in patients who undergo surgery for any reason. A fetal pelvic or ovarian cyst may predispose to torsion and subsequent auto-amputation either in utero or in the post natal period. Most patients are asymptomatic with an antenatal pickup and can be a cause for concern. Expectant management is advocated for smaller cysts (<4 cm) but surgery (laparoscopy or laparotomy) is diagnostic and curative.

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Auto-amputated Ovarian Cyst (AOC) in infants and/or the finding of an absent fallopian tube or ovary is rare and is probably due to torsion of the cyst pedicle[1]. The auto-amputated structure may then reattach to another surface or often become free floating, possibly with calcifications. We present our experience of 3 cases from three tertiary care hospitals and a review of the literature.

Materials and Methods

A systematic search of the databases (PubMed, Medline, Scopus, Google scholar) was performed using MeSH terms: neonate, infant, ovary and auto-amputation. The search was expanded by entering other terms (eg, ovarian surgery, torsion, child) to check for any missing article. The 'related articles' search facility in PubMed and references contained within relevant reports were also assessed as appropriate. We report three cases along with the literature review.

Results

Case Reports :

Case 1 : An 11-month-old girl presented with antenatal diagnosis of right Multicystic Dysplastic Kidney (MCDK) and normal left kidney. Follow up ante-natal Ultrasonography (US) showed increase in the size of the cyst. At 33 wk scan, the diameter of the cyst was 4.3 cm. In post-natal US, the size increased to 6 cm cyst. As the scan was suggestive of MCDK, initially a conservative management was planned. US at 10 months showed further increase in the size of the cyst to 7x6x4cm cyst with septa, fluid filled levels and some solid tissue, but no renal tissue. The cyst was occupying mainly right side of abdomen. Uterus and ovaries were not seen. MAG3 scan showed no function in the right kidney. Blood investigations including tumour markers were normal Urine culture had shown *E Coli* infection, which was treated with nitrofurantoin. With a provisional diagnosis of right kidney cyst, a laparoscopic removal of cyst was planned through transperitoneal route. To our surprise, a large auto-amputated free lying cyst (10x10x12 cm), having 500ml fluid with debris was present in the pelvis. The cyst was removed laparoscopically after aspirating the fluid. Histopathology revealed it as a benign ovarian cyst.

Case 2 : This 10-month-old infant was diagnosed to have a cyst on antenatal US. Post-natal US revealed a cyst (6x4cm) in pelvis with absent right ovary. The cyst was not resolving and appeared complex on US (thick walled with debris). Hence, an operative removal was planned. Laparoscopy revealed the ovary was absent on that side, other side ovary was normal looking, and the cyst to be free-floating lying-in pelvis. The cyst was removed laparoscopically. Histopathology revealed it as a benign ovarian cyst.

Case 3 : A one-day-old female neonate, born at 38+1-week gestation via a normal vaginal delivery, was transferred to a tertiary children's hospital with an

Editor's Comment :

- The routine 20-week anomaly antenatal ultrasound scans have led to an increased detection of fetal ovarian cysts. Although most of them (<4cm) may regress spontaneously, some may grow into large cysts and undergo torsion followed by auto-amputation. However, pre- and post-natal scans may fail to identify autoamputation and findings of a free-floating amputated cystic mas in association with absence of fallopian tube and/or ovary are important clues. Laparoscopy is a safe and effective tool for the diagnosis and removal of such ovarian cysts in neonates and infants.

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antenatal scan at 35+2 weeks showing a cystic lesion in the left side of the abdomen extending into the fetal pelvis. A fluid level and some debris were noted within the cyst, raising a suspicion of a mesenteric or a duplication cyst. An abdominal X-ray done after birth was unremarkable. The postnatal scan confirmed a 3X3cm cyst in the left side of the abdomen, filled with fluid and some debris. Apart from this finding, the neonate was otherwise well. She opened her bowels normally. Her blood investigations were normal. With a strong suspicion of a mesenteric or duplication cyst, baby underwent laparotomy which revealed a 6X6cm cyst adherent to the mesentery of the descending colon. It was noted to have a stump which was torted by 540 degrees and had a band leading on to the pelvis. Further examination revealed an absent left ovary with the left fallopian tube ending in a blind stump. The right ovary, tube and uterus were normal. The cyst was resected in total. The histopathology was consistent with an AOC with no features of malignancy.

Review of Literature :

In addition to the 3 cases reported by us, our review of literature identified that in the last 34 years (1980-2013) search in English literature, a total of 20 cases have been reported of infants (<1 year) with AOC. Three cases had a mobile abdominal mass was felt preoperatively. 4 cases were >5 cm in diameter. Only three of those cases were diagnosed as auto-amputated preoperatively. One infant had bilateral AOC, in which the histopathology showed hemorrhagic cysts of ovaries with dystrophic calcification, presumably related to intrauterine ischemic or hemorrhagic infarction. In 4 cases, initially a conservative approach with US follow up was undertaken, which showed no features of spontaneous regression, and finally operative removal was planned. Only 6 cases were operated laparoscopically and the other 14 cysts were removed by laparotomy. Interestingly, no malignancy was reported in any of these cases. More details are summarised in Table 1[1-3,6,13-25].

Discussion

AOC in infants was first reported in 1981[2]. The diagnosis and management have not been clearly established due to its rarity.

Clinically, AOC can be anticipated in cases presenting with freely mobile abdominal masses or cysts with variable locations in US[3]. Like ovary torsion, intracystic

Table 1 — *Showing auto-amputated ovarian cysts in infants (updated from reference 2)*

Author (reference)	Age at diagnosis	Pre-op. diagn.	US features	Size (cm)	Spontaneous regression	Op	Pathology
Koike, *et al*[1]	30-week gestation	No	Fluid debris level	3.2 x2.0	No	LT	Necrosis, hemorrhage, autolysis, calcification
Kennedy, *et al*[2]	5 months	No	Unknown	4.0 x 3.0	Unknown	LT	Calcified fibrous tissue
Kennedy, *et al*[2]	2 weeks	No	Unknown	3.2x1.5x2.5	No	LT	Calcification and ovarian stroma & follicles
Kuwata, *et al*[3]	28-week gestation	No	Side change of ovarian mass from left to right	4x3.5	No	LP	Necrosis with small amount of ovarian tissue
Brandt, *et al*[6]	3 months	No	Calcification	6.0	Unknown	LT	Unknown
Brandt, *et al*[6]	34-week gestation	No	Fluid debris level	3.0	Unknown	LT	Unknown
Jawad, *et al*[13]	41-week gestation	Yes	Unknown	4.2 x 3.7	No	LP	Necrosis, autolysis with calcification
Corbett, *et al*[14]	Day 1	No	Septae	3.5 x 2.5	Unknown	LT	Hemorrhagic ovary with calcification
Avni, *et al*[15]	Day 1	No	Fluid debris level	4.0	No	LT	Necrosis and fibrotic walls
Alrabeeah, *et al*[16]	38-week gestation	No	Unknown	Unknown	Unknown	LT	Necrotic tissue
Mordehai, *et al*[17]	30-week gestation	No	Fluid debris level	6.0 x 6.0	Unknown	LT	Unknown
Aslam, *et al*[18]	34-week gestation	No	Fluid debris level	3.0	No	LT	Necrotic tissue
Decker, *et al*[19]	Antenatal	No	Fluid debris level Hemorrhage	4.0 x 3.0	No	LP	Necrosis, autolysis with calcification
Tseng, *et al*[20]	Unknown	No	Unknown	Unknown	Unknown	LP	Unknown
Tsobanidou, *et al*[21]	27-week gestation	No	Fluid debris level	5.0	No	LT	Ovarian tissue with necrosis, hemorrhage, calcification
Amodio, *et al*[22]	37-week gestation	No	Fluid debris layer, septa, calcification	5x4x3	No	LT	Hemorrhagic, infarction, calcification
Zampieri, *et al*[23]	32-week gestation	yes	Free floating abdominal cyst	5.2x6	No	LP	Necrosis, no ovarian tissue
Zampieri, *et al*[23]	34-week gestation	Yes	Free floating abdominal cyst	3.5x4.5	No	LP	Hemorrhagic infarction of ovary
Visnjic, *et al*[24]	32 weeks gestation	No	Cyst with thick solid wall	4.5cm	No	LT	Hemorrhagic necrosis, no ovarian tissue
Dueck, *et al*[25]	37 weeks	No	Cystic abdominal mass	7x6cm	No	LT	Partly canalized and partly atretic L fallopian tube

LT - Laparotomy; LP - Laparoscopy; Op - operation; Preop - preoperative, Diag - diagnosis

hemorrhage and wall calcification is also found in ovarian auto-amputation, which suggests that auto-amputation could be a possible long-term consequence of torsion during fetal life[1]. Incidence of intracystic hemorrhage and torsion in ovarian cyst has been reported to be between 34% and 45%[4,5] and the risk of torsion increases with increasing size of cyst[6]. Hence, many authors have recommended surgical removal of cysts of more than 5 cm in diameter[7-9].

Our case 1 was unique, as it was the largest auto-amputated ovary cyst reported so far (10x10x12 cm), having 500ml fluid with debris. Other 7 cases, removed laparoscopically were smaller (up to 4cm in maximum diameter). We believe that size of the cyst should not be a contraindication of laparoscopic approach. As none of the cases in literature have shown malignancy, probably aspiration of a large cyst may make the operative procedure easier. Association of renal anomalies with ipsilateral Mullerian anomalies has been described in literature[9], but the association of renal anomalies with ovarian auto-amputation has never been reported. The association of right sided MCDK with ipsilateral auto-amputation of ovary could have embryological etiology, but it is difficult to prove. This association mislead us initially towards conservative treatment, otherwise we could have operated this baby earlier and this could be a learning lesson for future.

In a literature review, Ushakov, *et al* reported 25 cases of ovarian teratoma of greater omentum in adult. It was believed that auto-amputation and reimplantation of an ovarian dermoid cyst was the most common etiology of omental teratomas in those cases[11]. In a large literature review, Marshall reported 45 cases of ovarian enlargement in the first year of life (29 in the newborn and 16 in the remainder of the first year). Newborn ovarian lesions consisted of benign cysts of germinal or Graafian epithelial origin and granulose cell tumors, but one malignant lesion, a granulosa cell carcinoma, was reported from a premature stillborn. The infant ovarian lesions consisted of benign cysts, benign cystic teratomas, granulosa cell tumors and a single malignancy, a mesonephroma. But none of them were reported to be auto-amputated[12].

Conclusion

Laparoscopic removal of AOC is feasible and safe. No malignancy in AOC has been reported so far, which is encouraging for aspiration of large cysts during laparoscopic removal. However, as ovarian malignancy is known in even in this age group close observation and early intervention is required in these patients.

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