Ovarian Autoamputation in a Neonate: Case Report

Yenidoğanda Over Otoamputasyonu

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ABSTRACT This case was an asymptomatic abdominal cyst detected 3.5cm diameter in 38 gestation week fetus on antenatal ultrasonography. She was delivered by vaginal birth at 40th week. Postnatal USG revealed a cystic mass measuring 4x2,5 cm in the right pelvis containing an intracystic hypoechoic lesion with round calcification of the cyst wall. MRI image showed 4x5 cm diameter a hyperintense area, indicating hemorrhagic content. Autoamputated right ovarian cyst was found and removed from the abdomen. The cyst was surgically removed to confirm the diagnosis and treatment. In this article it was presented with diagnosis of necrosis due to auto-amputation in fetal ovarian cyst and it was surgically treated in neonatal period.

Key Words: Ovarian cysts; infant, newborn; torsion abnormality

ÖZET Otuz sekizinci gebelik haftasında yapılan antenatal ultrasonografisinde 3,5cm boyutunda asemptomatik abdominal kist tespit edilen olgu; 40. gebelik haftasında vaginal doğum ile doğurtuldu. Postnatal yapılan batın ultrasonografisinde sağ pelviste 4x2,5 cm çapında hipoekoik ve duvarında yuvarlak kalsifikasyonlar içeren lezyon saptandı. Pelvik MRI'da 4x5 cm çapında hiperintens hemorajik içerikli lezyon görüldü. Cerrahi sırasında otoampütasyona uğradığı tespit edilen kist tanı ve tedavi amacıyla çıkarıldı. Bu yazıda intrauterin dönemde tanı alan ve doğum sonrasında otoamputasyona bağlı nekroz gelişen ve cerrahi olarak tedavi edilen fetal over kisti olgusu sunulmuştur.

Anahtar Kelimeler: Over kistleri; bebek, yenidoğan; torsiyon anormalliği

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Fetal hormonal environment may cause the development of cysts in the ovaries in the intrauterine period. Development of pathologic cysts is thought to be caused by improper hormonal stimulation followed by unhealthy folliculogenesis, however most of the cysts regress in the postnatal period once the influence of maternal hormones has ceased.[1] Although most of these cysts are small simple cysts that resolve spontaneously after birth, larger cysts may undergo torsion and autoamputation.[2] Autoamputation of ovarian cysts in infants under 1 year of age is a rare complication, and most likely follows a severe degree of cystic torsion with subsequent ischemia and necrosis of the entire adnexal structures or the cyst pedicle.[3] Clinically, autoamputated gonadal lesions or ovarian cysts may present as freely mobile abdominal masses on ultrasound (USG).[4]

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We report a neonate with an autoamputated ovarian cyst, and present a review of the reported cases in infants under 1 year of age.

CASE REPORT

A female fetus presented with an abdominal cystic mass measuring 3.5 cm on antenatal USG at 38 weeks of gestation. The baby was born vaginally at the end of 40 weeks of gestation with a birth weight of 3.2 kg. Physical examination was unremarkable, there was no palpable mass in the abdomen. Hematologic and biochemical investigations were normal. Postdelivery USG revealed a 4x2.5 cm cystic mass containing an intracystic hypoechoic lesion with round calcification of the cyst wall in the right pelvis. In magnetic resonance imaging (MRI), T1-weighted fat saturation image showed a hyperintense area 4x5 cm in diameter, indicating hemorrhagic content (Figure 1) The neonate underwent elective laparotomy on the third day after birth. During surgery, a free-floating, brown, round, smooth cystic lesion measuring 3.8 cm in diameter, without a stalk or peritoneal attachment, was found in the peritoneal cavity and removed. The right ovary was absent with a blind end of the right fallopian tube. The cyst was filled with a wine-colored fluid. Histopathologically, the tumor mostly consisted of extensive hemorrhagic necrotic autolytic tissue with dystrophic calcification, but did not contain any ovarian tissue. The postoperative course was uneventful, and the patient was discharged from hospital on the postoperative seventh day.

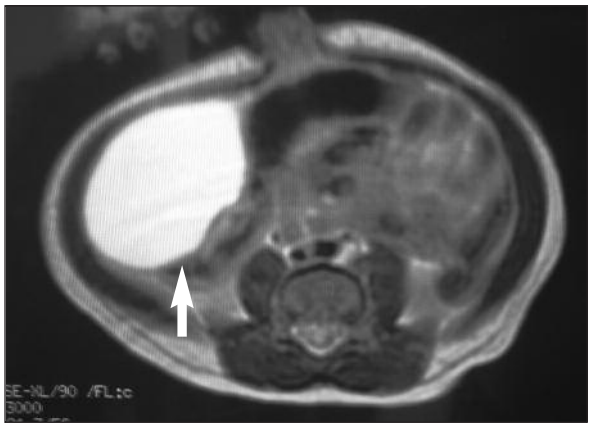

FIGURE 1: Postnatal abdominopelvic axial T1- weighted magnetic resonance imaging showing a hyperintense area 4x5 cm in diameter.

DISCUSSION

Ovarian cysts have a wide spectrum of pathology ranging from functional (non-neoplastic) ovarian cysts to ovarian torsion and from benign to highly aggressive malignant neoplasms.[5] Most ovarian cysts in infants are follicular cysts that are thought to result from excessive stimulation of the fetal ovaries by human chorionic gonadotropin, and are frequently seen in the offspring of mothers with diabetes mellitus, Rh isoimmunization and toxemia.[3] Nussbaum et al. classified the USG patterns of ovarian cysts into simple and complex, based on the pathologic findings from a series of affected infants.[3] Fetal ovarian cysts are often diagnosed by USG during antenatal period. During infancy, the consensus on the management of simple ovarian cysts less than 5 cm in diameter includes a wait-and-see approach with USG monitoring, since they tend to regress spontaneously.[6] Spontaneous resolution of cysts <3 cm is not uncommon both prenatally and after delivery. Some suggest, however, aspiration of the cystic contents is possible even in ovarian cysts exceeding 4 cm in diameter. However, some authors advocate obstetric procedures, such as intrauterine puncture of the cyst and induction of labor.[7] In cases of simple cysts larger than 5 cm in diameter and complex cysts, surgical removal of the mass is most often recommended.[8] Kennedy et al. first reported the autoamputation of ovarian cysts in infants under 1 year of age, and noted that it is extremely rare.[8] Its diagnosis and treatment have not been clearly established due to its rarity. There have only been 13 reported cases of autoamputated ovarian cysts in infants under 1 year of age including our case, and only one of these cases was diagnosed as an autoamputated ovarian cyst preoperatively.[9] Although the diagnostic hallmark of autoamputated cysts is the detection of a freely mobile mass in the abdominal cavity, 12 cases were free of clinical symptoms. In the remaining one case, a mobile abdominal mass was detected preoperatively on physical examination.[6] Only two cases has cysts larger than 5 cm in diameter, which is the suggested diameter for surgical removal of ovarian cysts in infants.[6,7] The USG features were almost the same as a complicated

ovarian cyst pattern.[10] Four of the reviewed cases adopted a wait-and-see approach with USG monitoring, but all of them eventually underwent surgical removal due to the absence of spontaneous regression. Postnatal symptomatic cysts or cysts with a diameter greater than 5 cm that do not regress or enlarge should be treated, but uncomplicated asymptomatic cysts less than 5 cm in diameter should only be observed and reassessed by serial ultrasonography. If they regress spontaneously, no surgical intervention is necessary independent of their sonographic findings. Simple ovarian cysts may be treated with fenestration of the cyst or cystectomy. Complex cysts should be treated with cystectomy when possible to avoid unnecessary loss of functional ovarian tissue. In cases where no viable tissue remains, oophorectomy may be necessary. Judging from pathological features in this review, intracystic hemorrhage and wall calcification that can be seen in twisted ovaries which have not undergone amputation are also the critical features of ovarian autoamputation. These findings imply that autoamputation in neonate is one possible long-term consequence of torsion during fetal life and no more spontaneous regression regardless of cyst size. In only our case, intracystic hemorrhage was revealed preoperatively by MRI. Considering the difficulty of the preoperative diagnosis of ovarian autoamputation, MRI would be useful when there is no definitive information or spontaneous regression of cyst examined by consecutive USG examinations.

Twenty two cases of teratoma of the greater omentum have been reviewed. In some of these cases, autoamputated ovarian teratomas became reimplantated as omental masses. This finding suggests that autoamputated ovarian cysts may develop into omental masses and have malignant potential in the future. Taking this possibility and the feature of no spontaneous regression into consideration, early surgical removal of an autoamputated ovarian cyst is preferable to take a wait and see approach.

In conclusion, in cases of ovarian cysts under 1 year of age with wall calcification and intracystic hemorrhage, we should keep the possibility of ovarian cyst autoamputation in mind. Absence of spontaneous regression in USG, intracystic hemorrhage, wall calcification in MRI and detection of a typical wandering tumor are useful hints for diagnosing autoamputation.

Although neonatal simple ovarian cysts are rare, they may cause abdominal distension, and other complications. Furthermore, if these findings are detected preoperatively, one should strongly suggest that such type of ovarian cyst will no longer undergo spontaneous regression and surgical removal may be the way to go.

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