ELSEVIER

JMIG The Journal of Minimally Invasive Gynecology

Case Report

Laparoscopic Management of Autoamputated Ovary in Newborns: A Report of 2 Cases

Hannah N. Ladenhauf, MD*, Martha G. Brandtner, MD, Mircia A. Ardelean, MD, Christa Schimke, MD, and Roman Metzger, MD

From the Department of Pediatric and Adolescent Surgery, Paracelsus Medical University, Salzburg, Austria (all authors).

ABSTRACT Intrauterine autoamputation of the ovary is an extremely rare diagnosis in the pediatric population. The current literature is limited to contradictory recommendations, while a standard management protocol for autoamputated adnexa secondary to intrauterine ovarian torsion is yet to be established. We report 2 cases of auto-amputation of the ovary, leading to a free-floating intra-abdominal cyst in the newborn. Laparoscopic management was successful in both cases. Journal of Minimally Invasive Gynecology (2017) ■, ■–■ © 2017 AAGL. All rights reserved.

Keywords: Adnexal cyst; Autoamputation ovary; Newborn

Ovarian pathologies are a rare finding in newborns [1–6]; however, advanced radiographic imaging techniques, including high-resolution ultrasound (US) as well as magnetic resonance imaging (MRI), facilitate the identification of intra-abdominal pathologies. In most cases, early detection and precise assessment is possible [7]. Benign cysts, autoamputated ovary and/or fallopian tube, teratomas, and mesenterial cysts or intestinal duplications constitute the most important differential diagnoses of pathologies of the ovary and fallopian tubes.

Autoamputation of the ovary is defined as a free-floating tubo-ovarian remnant that is dislocated from its original anatomic position. The uterus, contralateral ovary, and adnexa, as well as the urinary tract system, are not involved [1]. The amputated ovary may undergo resorption, calcify, or become a cystic lesion, which can present as a free-floating intra-abdominal mass without connection to other organs. Newborns with autoamputated ovary are often asymptomatic and are diagnosed because of a palpable mass in the lower abdomen. US including Doppler sonography and subsequent MRI studies may increase suspicion of autoamputation of the ovary and can provide additional information to rule out differential diagnoses.

Surgical treatment is described in most cases, with most centers performing laparotomy for resection of the remnant structures. Conservative treatment may be successful in selected cases, however [2].

We present 2 cases of autoamputation of the ovary, in 1 case including the adnexa, both of which were successfully treated with laparoscopic removal of the ovarian remnant.

Case 1: Autoamputation of the Left Fallopian Tube and Ovary

A 9-day-old girl with a prenatally diagnosed intra-abdominal cystic lesion of 4.4 × 3.8 cm at 36 weeks of gestation was referred to our clinic. The ovaries could not be identified; however, no further abnormalities were detected. The prenatal period and birth had been uneventful. US follow-up after 1 week revealed a clear-cut cystic lesion of 5 × 2.8 × 4.5 cm containing hypoechoic and hyperechoic fluid with sedimentation as part of the left ovary (Fig. 1A). In addition, the left ovary contained a small follicle 4 mm in diameter. The infant was asymptomatic, and no abdominal mass was palpable. Tumor markers (alpha fetoprotein and human chorionic gonadotropin) were within normal ranges. The follow-up US after 1 month showed the cyst in the right lower abdomen and was described as part of the right ovary. The right ovary contained several

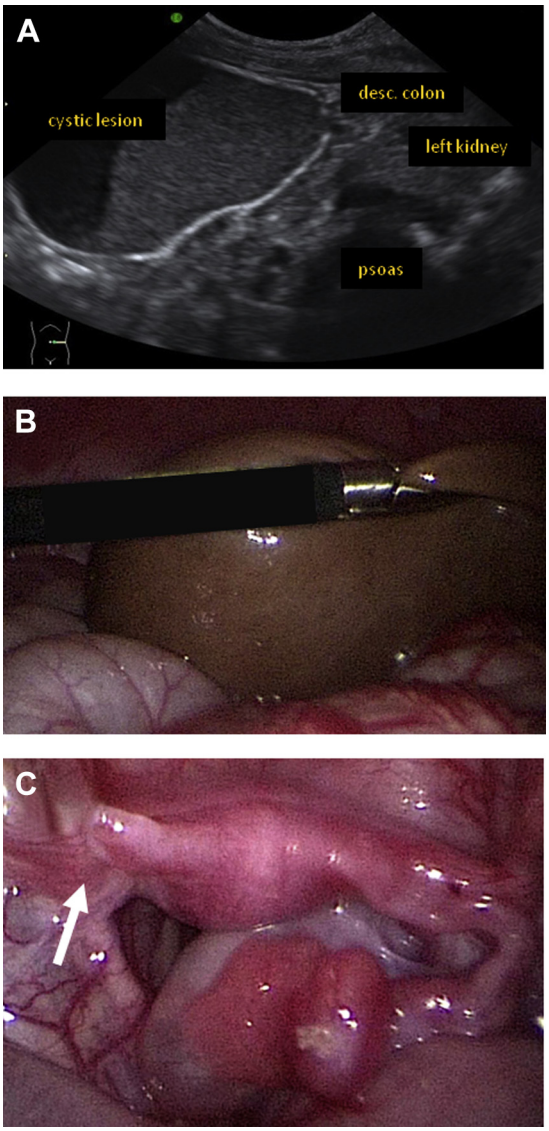
The authors declare that they have no conflicts of interest.
Ethics approval: According to the regulations of the local ethics committee, this study was exempt from review due to the retrospective character of this data analysis. However, written parental consent was obtained in both cases.
Corresponding author: Hannah N. Ladenhauf, MD, Muellner Hauptstrasse 48, 5020 Salzburg, Austria.
E-mail: h.ladenhauf@salk.at

Submitted January 31, 2017. Accepted for publication April 14, 2017.
Available at www.sciencedirect.com and www.jmig.org

1553-4650/$ - see front matter © 2017 AAGL. All rights reserved.
http://dx.doi.org/10.1016/j.jmig.2017.04.006

Fig. 1

(A) US showing a clear-cut cystic lesion of 5 × 2.8 × 4.5 cm with hypoechoic and hyperechoic fluid and sedimentation interpreted as part of the left ovary. **(B)** Intraoperative puncture of the cyst. **(C)** Autoamputation of the left ovary and fallopian tube. The arrow indicates the fallopian stump.



follicles 6 to 10 mm in diameter. Tumor markers were still negative, and the infant remained asymptomatic.

At age 2 months, exploratory laparoscopy using one 5-mm and two 3-mm umbilical ports was performed. A free-floating brownish cyst was identified in the umbilical region of the abdomen. The right ovary presented in its typical location and was adequately suspended via the broad and ovarian ligaments. The left ovary could not be identified, and the left fallopian tube (Fig. 1C, arrow) terminated blindly approximately 1 cm from the uterus, indicating autoamputation not only of the left ovary, but also of the fallopian tube (Fig. 1C). The cyst was punctured, and 50 mL of turbid red-brownish fluid was aspirated (Fig. 1B). The cyst was then removed via an extraction bag. Pathological evaluation revealed a cystic lesion with ovarian stroma and parenchyma with single oogonia at the margin. No signs of teratomatous elements or malignancy were seen. An endocrinology workup was scheduled, and Turner's syndrome was ruled out.

Case 2: Autoamputation of the Right Ovary

At 36 weeks of gestation, a patient was referred for pediatric surgical counseling owing to a persistent cystic lesion of the right ovary measuring 5 × 3 cm. At age 7 days, the cyst was 5.9 × 4.9 × 2.3 cm in size. No other pathologies were identified. The cyst was described as clear-cut, containing hypoechoic and hyperechoic fluid (Fig. 2A). The cyst was first seen in the left and later in the right lower abdomen. The left ovary did not show signs of pathology; however, the right ovary could not be identified. The infant was asymptomatic, with no palpable mass.

The cyst decreased in size over time, but at age 4 months still measured 3.8 × 3.6 × 3.6 cm, and the exact origin remained unclear. Subsequent MRI at age 6 months revealed a 4 × 3.4 × 3.4 cm cystic hemorrhagic lesion with an air-fluid level and peripheral enhancement, consistent with a polypoid lesion. A teratoma was less likely, owing to the absence of fat tissue. Levels of tumor markers (alpha fetoprotein and human chorionic gonadotropin) were within physiological ranges. The left ovary appeared normal. A torsion of the right ovary was suspected.

At age 6 months, an exploratory laparoscopy was performed, using one 5-mm and two 3-mm umbilical ports. A free-floating brownish cystic lesion was detected and punctured (Fig. 2B). Following the right fallopian tube (Fig. 2C, arrow), the tube was found to be intact, but an ovary could not be identified (Fig. 2C). The left ovary appeared on the left side of the pelvis and was adequately suspended via the broad and ovarian ligaments. The cyst was punctured and removed via an extraction bag. A pathological workup ruled out teratoma and malignancy. The postoperative course and 11-month follow-up were uneventful.

Discussion

Autoamputation of the ovary and/or fallopian tube is a very rare diagnosis in the pediatric population. The most widely accepted theory to explain its etiology seems to be chronic adnexal torsion and subsequent devascularization. Whether embryologic factors may be involved remains unknown. Possible risk factors may include anatomic alterations, such as malformation or elongation of the tuboovarian ligament. Any increase in weight of the ovary, most often caused by a cystic lesion, may lead to ovarian torsion. Ovarian stimulation, as occurs during pregnancy, may lead to increased size and weight of the ovary and may predispose to ovarian torsion [8]. Rare cases of ovarian