

# The Rare Coexistence of Isolated Unilateral Adnexal Agenesis and Microperforated Hymen: A Case Report

## İzole Unilateral Adneksiya Agenesisi ile Mikroperfore Hymen Tanılarının Nadir Birlikteliği: Olgusu

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### ABSTRACT

We share an 18-year-old patient with coexistence of isolated unilateral adnexal agenesis and microperforated hymen (MH). The rare type of partially obstructive congenital anomaly is MH. If the menstrual products or vaginal secretions are not fully evacuated from the vagina, the retained blood may become infected and lead to pyocolpos, tuboovarian abscess or systemic infection. Giant pyocolpos complicates the assessment of adjacent anatomical structures. In imaging methods, tuboovarian abscess-like appearance may occur. On laparoscopic observation; while describing the unilateral absence of the ovary and fallopian tube, we did not encounter any other anatomical malformations. Peroperatively, we excluded the prediagnosis of tuboovarian abscess and diagnosed pyocolpos. Mucopurulent fluid was drained by hymenotomy. No complications were observed in the postoperative six-month follow-up.

**Keywords:** Adnexal agenesis; microperforate hymen; pyocolpos; tuboovarian abscess

### ÖZET

Bu makalede, 18 yaşında izole unilateral adneksiya agenezi ve mikroperfore hymen (MH) tanılarının birlikte görüldüğü hasta sunulmaktadır. MH, kısmi obstrüktif konjenital anomali türüdür. Menstrüel sıvı veya vajinal sekresyonlar vajinadan tam olarak boşaltılmazsa, geride kalan artıklar enfekte olabilir. Bu durum pyokolpos, tuboovaryan abse veya sistemik enfeksiyona yol açabilir. Dev piyokolpos, komşu anatomik yapıların değerlendirilmesini zorlaştırabilmektedir. Görüntüleme yöntemlerinde, tuboovaryan abse benzeri görünüm oluşabilmektedir. Laparoskopik gözlemede; unilateral adneksiya agenezi saptandı. Ek anatomik malformasyon izlenmedi. Peroperatif olarak tuboovaryan abse ön tanısını dışlandı ve piyokolpos tanısı koyuldu. Mukopürülan sıvı hymenotomi ile boşaltıldı. Ameliyat sonrası altı aylık takipte herhangi bir komplikasyon görülmedi.

**Anahtar Kelimeler:** Adneksiya agenezi; mikroperfore hymen; piyokolpos; tuboovaryan abse

Hymen is a localized membrane on the distal side of the vagina. Its canalization is completed just before birth.<sup>1</sup> Cervical gland secretions and menstrual blood products are excreted through this orifice. If this canalization is interrupted, hymen remains partially obstructed.<sup>1</sup> The rare type of partially obstructive congenital anomaly is microperforated hymen (MH).<sup>2</sup> Its exact incidence has not been reported in

the literature. Cases or case series are reported in the literature.<sup>2</sup> Due to insufficient data, the management of this congenital anomaly is controversial.<sup>1</sup>

Light and irregular menstrual periods can be considered a healthy condition during adolescence. Therefore, the diagnosis of MH can be delayed until the late adolescent period. If the menstrual products or vaginal secretions are not fully evacuated from the

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vagina, the retained blood may become infected and lead to pyocolpos, tuboovarian abscess or systemic infection.<sup>3,4</sup> This septic condition may be the result of MH. Giant pyocolpos complicates the assessment of adjacent anatomical structures. In imaging methods, tuboovarian abscess-like appearance may occur.

MH is usually an isolated anomaly.<sup>5</sup> It is rarely associated with uro/ano genital anomalies.<sup>5</sup> Unilateral agenesis of the ovary and/or uterine tube accompanying MH is an extremely rare condition.<sup>6</sup> Three isolated cases of unilateral ovarian agenesis have been reported in the literature.<sup>7</sup> In this study, we share the coexistence of isolated adnexal agenesis and MH.

## CASE REPORT

An 18-year-old girl was admitted to the emergency department with complaints of abdominal pain and subfebrile fever for one week. The patient was febrile (38.5°C). No pathological finding was detected on the abdominal examination. On the gynecological examination, there was an appearance compatible with microperforated hymen. She was virgin. The age of menarche was 12 years. She had irregular menstrual cycle and no previous abdominal surgery. In laboratory examination, serum white blood cell level was found to be  $10.5 \times 10^3/\text{mm}^3$ , and c-reactive protein (CRP) 119 mg/L. Radiological abdominal ultrasonography reported a mass of 11 cm originating from the right adnexa and thought to be a tuboovarian abscess. Then, in the computed tomography (CT) examination; the uterus was normal, a finding similar to the ultrasonography in terms of the right adnexa was described, while the left adnexal area could not be clearly evaluated (Figure 1). She was interned in the gynecology service. Intravenous cephalosporin and metronidazole combination therapy was planned. During the 48-hour follow-up, the patient's fever progressed, abdominal pain intensified, and diagnostic laparoscopy was performed. Unexpectedly, right tuboovarian abscess formation was not observed. The external contour of the uterus, the right round ligament, right tuba uterine and right ovary was normal. While the left round ligament was normal, the left ovary and left tuba were not observed. No residual formation was observed in the left adnexal area. The bilateral ureters were in their normal anatomical positions (Figure 2).

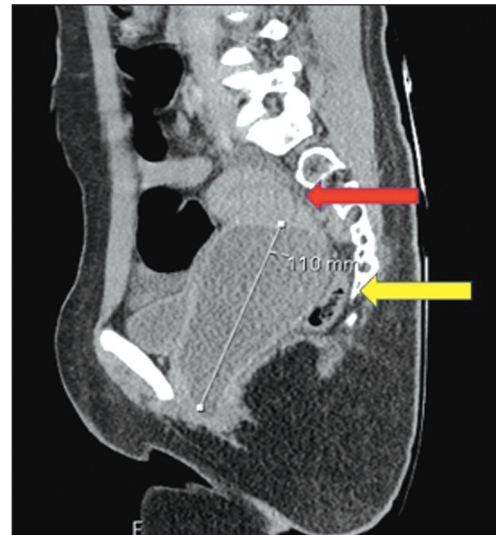


FIGURE 1: Preoperative CT; yellow arrow shows giant pyocolpos mimicking tuboovarian abscess, red arrow shows uterus.

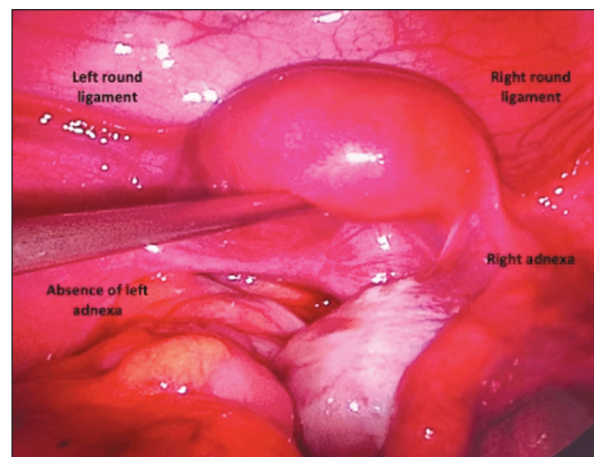


FIGURE 2: Absence of peroperative unilateral adnexa.

Then, digital rectal examination was performed and infective vaginal discharge began to leak from the microperforated hymen. Pyocolpos was diagnosed. Following the hymenotomy with a cross incision, 200 cc of mucopurulent fluid was drained. Vaginal walls and single cervix were normal anatomically on vaginoscopy. Hymenoplasty was performed. She was followed up in the service for 48 hours postoperatively. Peroral antibiotics were prescribed and she was discharged. No additional gynecological, urinary, or anorectal anomalies were detected in postoperative magnetic resonance imaging (MRI) examination. Basal hormone levels evaluated in the

postoperative menstrual cycle; FSH (6.04 mIU/ml), LH (3.97 mIU/ml), TSH (2.8 mIU/l), prolactin (7.6 ng/ml), estradiol (55.8 pg/ml), total testosterone (14.9 ng/ml), free testosterone (0.9 pg/ml), DHEA-S (197.6 ng/ml), serum hormone binding globulin (53.8 nmol/L), 1.4 delta androstenedion (0.8 ng/mL), AMH (3.8 ng/mL) were normal. Genetic analysis confirmed 46 XX karyotypes. No stenosis was observed in the hymenal ring during postoperative six-month follow-up. It was found that menstrual blood flow was normal and there was no vaginal discharge. Informed consent was obtained from the patient.

## DISCUSSION

The clinical symptoms of MH, which is a partially obstructive hymenal anomaly, may differ in each patient.<sup>1</sup> The clinical presentation depends on the diameter of the hymen orifice.<sup>1</sup> MH cases can be asymptomatic for life. However, these patients may experience symptoms of dyspareunia, dysmenorrhea and severe abdominal pain during adolescence.<sup>4</sup> In these women with regular or irregular menstruation, the diagnosis of MH may be delayed until late adulthood.<sup>1</sup> In MH cases, pyocolpos occurs as a result of stasis of vaginal discharge due to partial obstruction of the hymen. Pyocolpos is a surgical emergency. If pyocolpos is not treated urgently, it can lead to vesicovaginal fistulae, destruction of the vaginal mucosa, and sepsis.<sup>4,5</sup> In addition, it should be considered that giant pyocolpos can be interpreted as tubaovarian abscess in preoperative imaging methods. Perioperatively, we excluded the prediagnosis of tubaovarian abscess and diagnosed pyocolpos. On laparoscopic observation; while describing the unilateral absence of the ovary and fallopian tube, we did not encounter any other anatomical malformations. Notably our

patient had no history of previous abdominal surgery, no residual structure from the autoamputated adnexa in the pelvic area, and no additional anatomical anomaly. The absence of one or both uterine tubes and ovaries is extremely rare. Mylonas et al. reported three cases of unilateral ovarian agenesis.<sup>7</sup>

A comprehensive genital examination should be performed in adolescent patients presenting with abdominal pain and irregular menstrual cycle. It should be kept in mind that MH cases may show clinical findings of giant pyocolpos mimicking tubaovarian abscess. Isolated unilateral adnexal agenesis and MH are very rare diagnoses in the literature. The coexistence of these diagnoses is the most important element that makes our study valuable.

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*During this study, no financial or spiritual support was received neither from any pharmaceutical company that has a direct connection with the research subject, nor from a company that provides or produces medical instruments and materials which may negatively affect the evaluation process of this study.*

### Conflict of Interest

*No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.*

### Authorship Contributions

**Idea/Concept:** Onur Yavuz; **Design:** Onur Yavuz; **Control/Supervision:** Onur Yavuz, Mehmet Güney; **Data Collection and/or Processing:** Onur Yavuz, Begüm Ertan; **Analysis and/or Interpretation:** Onur Yavuz, Mehmet Eyüphan Özgözen; **Literature Review:** Onur Yavuz, Aslı Akdöner; **Writing the Article:** Onur Yavuz, Begüm Ertan; **Critical Review:** Onur Yavuz, Mehmet Güney.

## REFERENCES

1. Tardieu SC, Appelbaum H. Microperforate Hymen and Pyocolpos: A Case Report and Review of the Literature. *J Pediatr Adolesc Gynecol.* 2018;31(2):140-2. [[Crossref](#)] [[PubMed](#)]
2. Watrowski R, Jäger C, Gerber M, Klein C. Hymenal anomalies in twins--review of the literature and case report. *Eur J Pediatr.* 2014;173(11):1407-12. [[Crossref](#)] [[PubMed](#)]
3. Stewart JF. Recurrent urinary tract infection in the female child secondary to a partial imperforate hymen. *J Urol.* 1970;103(3):353-6. [[Crossref](#)] [[PubMed](#)]
4. Sanfilippo AM, Mansuria SM. Microperforate hymen resulting in pelvic abscess. *J Pediatr Adolesc Gynecol.* 2006;19(2):95-8. [[Crossref](#)] [[PubMed](#)]
5. Winderl LM, Silverman RK. Prenatal diagnosis of congenital imperforate hymen. *Obstet Gynecol.* 1995;85(5 Pt 2):857-60 [[Crossref](#)] [[PubMed](#)]
6. Sivanesaratnam V. Unexplained unilateral absence of ovary and fallopian tube. *Eur J Obstet Gynecol Reprod Biol.* 1986;22(1-2):103-5. [[Crossref](#)] [[PubMed](#)]
7. Mylonas I, Hansch S, Markmann S, Bolz M, Friese K. Unilateral ovarian agenesis: report of three cases and review of the literature. *Arch Gynecol Obstet.* 2003;268(1):57-60. [[Crossref](#)] [[PubMed](#)]