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Incidental diagnosis of unilateral renal and adnexal agenesis in a 46-year-old multiparous woman

Authors' Contribution:
Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
Funds Collection G

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Patient: Female, 46
Final Diagnosis: Unilateral adneksial and renal agenesis
Symptoms: Menometrorrhagia
Medication: —
Clinical Procedure: Total abdominal hysterectomy and unilateral salphingoophorectomy
Specialty: Obstetrics and gynecology

Objective: Rare disease
Background: Unilateral renal and adnexal agenesis is quite rare. Absence of any uterine abnormality accompanying current urogenital abnormalities is even rarer.
Case Report: We report on the case of a 46-year-old multiparous woman, incidentally diagnosed to have unilateral renal, ovarian, and tubal agenesis just before hysterectomy due to menometrorrhagia and myoma uteri.
Conclusions: Any diagnosis of a urogenital abnormality necessitates investigation of comorbid renal or genital abnormalities.

Key words: agenesis • adnexal agenesis • renal agenesis • incidental • multiparity

Full-text PDF: <http://www.amjcaserep.com/download/index/idArt/883970>

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Background

We report on the incidental recognition of unilateral renal and adnexal agenesis in a 46-year-old multiparous patient during extended evaluation just before hysterectomy. Comorbid presence of adnexal and renal agenesis is quite rare. It is due to embryological development and is usually accompanied by uterine abnormalities, but this case had no uterine abnormality.

Case Report

A 46-year-old, gravidity 4, parity 4 patient was admitted to our clinic with meno-metrorrhagia of about 4 years duration. All of her 4 previous pregnancies were normal and resulted in normal vaginal delivery. Gynecological examination revealed a large uterus with distorted shape. Ultrasonography revealed a 10-cm possible leiomyoma originating from the corpus posterior part of the uterus, with a normal right ovary. We were unable to observe the left ovary. To clarify the diagnosis, an abdominopelvic CT was performed, confirming ultrasonographic findings, but also showing a confusing finding – the absence of a left kidney (Figure 1). Cervical smear and endometrial biopsy were performed to rule out any comorbid pathology, and were reported as normal cytology and irregular proliferative endometrium. After consultation with the Urology Department, an intravenous pyelography was done, revealing a normally functioning right kidney and an absent left kidney (Figure 2).

Intraoperatively, the uterus was observed as globally enlarged by an approximately 10-cm leiomyoma deforming the shape of the uterus. Also of interest, the left round ligament was intact but the left ovary and fallopian tube were not observed (Figure 3). A double J catheter was administered to the right ureter intraoperatively by cystoscopy as a precaution against any urethral damage. Total abdominal hysterectomy and right salpingo-oophorectomy was done. Pathological examination of

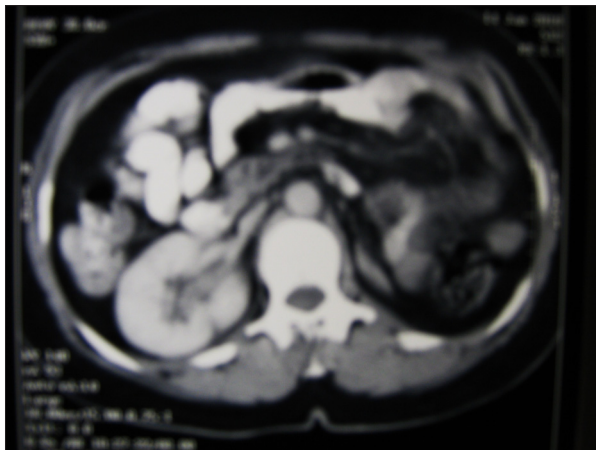


Figure 1. Abdominopelvic CT; absence of left kidney.



Figure 2. Intravenous pyelography, functioning right kidney and absence of left kidney.

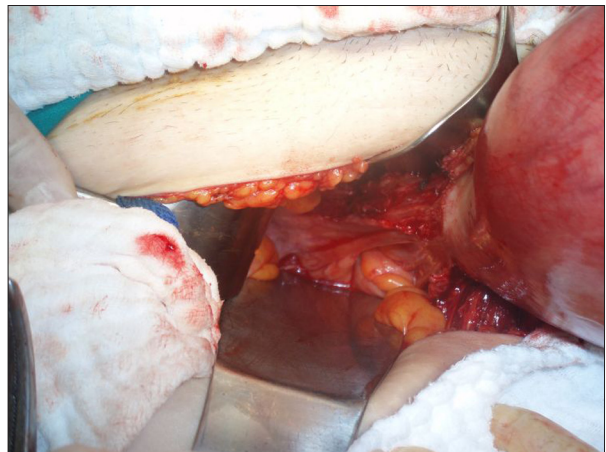


Figure 3. Intraoperative appearance of absence of left adnexa.

the specimen was reported as leiomyoma of the uterus, with a normal right ovary.

Discussion

Unilateral ovarian and fallopian tube agenesis has been explained by 2 mechanisms; (a) adnexal torsion at some time in life and (b) congenital absence [1]. In this case, the presence of another congenital abnormality – renal agenesis on the same side – primarily supports the second hypothesis as the etiology. Although agenesis of unilateral kidney is frequent (1 per 500–1000 autopsies and 1 per 2900–3200 births) [2], the suggested incidence of absence of unilateral adnexa is approximately 0.0089% (1 in 11 240 cases) [3]. Due to close embryological development, unilateral renal agenesis may be associated with other mesonephric and paramesonephric ductal

anomalies. Genital anomalies occur in 37–60% of females and 12% of males with congenital unilateral renal agenesis [4].

Acien et al classified urogenital abnormalities based on clinical and embryological development [5]. According to our Medline search, unilateral absence of kidney and adnexa is usually accompanied by uterine abnormalities and there are very few cases reported to have an isolated adnexal and renal agenesis without comorbid uterine abnormalities [6]. Although this case can be included in the Type 1 group, absence of any uterine abnormality is extraordinary.

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Conclusions

Urogenital abnormalities may be diagnosed at any time of life. The presence of urinary or genital abnormality should warn us about searching for any abnormality related with the other. Asymptomatic relatives of the subject should be screened because renal abnormality may be part of a familial syndrome [7]. Incidental diagnosis of such patients suggests that these congenital abnormalities do not disrupt female fertility, especially when not accompanied by uterine malformations.