Congenital interruption between the ampulla and fimbria of the fallopian tube: case report

Xiaotong Dong, MD; Shengrui Zhao, MM; Peng Liu, MM; Qian Yu, MD; Lei Yan, MD

We reported 5 patients with unilateral or bilateral tubal discontinuity between the ampulla and fimbria, occasionally detected through laparoscopy combined with hysteroscopy at the Reproductive Hospital Affiliated with Shandong University from 2017 to 2023. Three cases were observed to have this malformation on the left fallopian tube, 1 case on the right side, and 1 case bilaterally. None of these cases were combined with urological malformations. After surgery, there was 1 instance of postoperative delivery, 1 ongoing pregnancy, and 3 not yet conceived. The congenital ampulla and fimbria interruption of the fallopian tube may be independent of other significant deformities of reproductive or renal tracts. However, it can lead to hydrosalpinx and primary or secondary infertility. Surgical management options, such as salpingostomy, salpingectomy, and tubal ligation, have shown the potential to improve pregnancy outcomes.

Key words: case report, fallopian tube, infertility, laparoscopy, malformations

Introduction

The fallopian tubes are a crucial component of the female reproductive tract originating from the Müllerian ducts, which serve as a conduit connecting the ovary to the uterus and play an indispensable role in facilitating fertilization and supporting preimplantation embryonic development.[1] Problems, such as hydrosalpinx, inflammation, or anomalies of the fallopian tubes, can cause infertility and ectopic pregnancy. The congenital ampulla and fimbria discontinuity of the fallopian tube is rare and may be incidentally found during laparoscopy. We have identified 5 cases exhibiting this rare condition. In this report, we present a detailed analysis of one typical case while providing supplementary materials showcasing other cases. Prior informed consent was obtained from all patients.

Typical case

A 30-year-old nulligravida woman was admitted in August 2017 with 2 years of infertility. She reported regular periods every 30 days, and her husband's semen examination showed no significant abnormalities. There was no history of prior surgery or sexually transmitted diseases, and she tested negative for pathogens, including *Chlamydia trachomatis*, *Treponema pallidum*, *Neisseria gonorrhoeae*, HIV, and Mycobacterium tuberculosis. Preoperative hysterosalpingography (HSG) revealed hydrosalpinx on both sides (Figure, A). The patient underwent laparoscopy combined with hysteroscopy to treat hydrosalpinx on August 7, 2017. Hysteroscopy confirmed a normal-appearing uterine cavity, whereas laparoscopy identified bilateral fallopian tube pathology characterized by interruption of the ampulla and the fimbria, resulting in bilateral ampulla hydrosalpinx (Figure, B and C). No pelvic adhesions or signs of pelvic infection were observed during the surgery. The bilateral ovaries and the uterus appeared normal. Hydrotubation using methylene blue confirmed bilateral tubal obstruction at the distal end of the ampulla. With the consent of her husband, a salpingostomy on both ampulla was performed. Methylene blue was used after salpingostomy to confirm the patency of the bilateral tubes. After the surgery, because of personal reasons, she took contraception for several years and now conceived spontaneously for 6 months (Table).

Additional cases are detailed in the Supplementary Materials.

Discussion

The fallopian tube serves as the conduit connecting the ovary to the uterus and plays an essential role in supporting fertilization and preimplantation embryonic development.[2] In recent years, because of advancements in assisted reproductive technology (ART), the fallopian tube has been underestimated for its nonessential role in reproduction since the success of in vitro fertilization >30 years ago.

Tubal factors account for approximately 25% of infertility cases, with hydrosalpinx accounting for about 10% to 30% of these.[3] Pelvic inflammation is the most common cause of hydrosalpinx. It is now widely accepted that fluid within a hydrosalpinx significantly reduces pregnancy rates in ART, resulting in a 50% decrease in success

From the School of Medicine, Cheeloo College of Medicine, Shandong University, Jinan, China (Drs Dong and Liu); Institute of Women, Children and Reproductive Health, Shandong University, Jinan, China (Drs Dong, Zhao, Liu, Yu, and Yan); Shandong Key Laboratory of Reproductive Medicine, Jinan, China (Drs Dong, Zhao, Liu, Yu, and Yan)

X.D. and S.Z. are joint first authors.

The authors report no conflict of interest.

Institutional Review Board approval is acquired and provided with the attachment. Consent was obtained from the patient, and no personal information was disclosed.

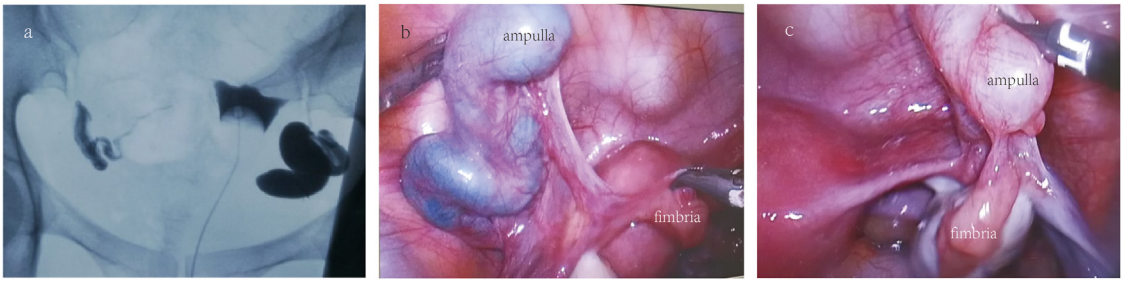
This paper was funded by BJHPA-2022-SHZHYXZHQNYJ-LCH-003 and Shandong Provincial Key Research and Development Program (2020ZLYS02).

Corresponding author: Lei Yan, MD.
yanlei@sdu.edu.cn

2666-5778/$36.00

http://dx.doi.org/10.1016/j.xagr.2024.100353

FIGURE
Pictures of a typical case



A, Left fallopian tube after hydrotubation with methylene blue. **B,** Right fallopian tube. **C,** Hysteroscopic image.

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compared with women without hydrosalpinx.[3] Salpingostomy, or distal tubal plastic surgery in the management of hydrosalpinx, is an alternative for women desiring natural conception, although ectopic pregnancy rates as high as 10% have been reported. A systematic review[4] comprising 22 studies and involving 2810 patients demonstrated that women who underwent salpingostomy for hydrosalpinx achieved a spontaneous pregnancy rate of 27%. In this article, we present 2 cases of salpingostomy with 1 spontaneous conception after surgery, suggesting that salpingostomy can be considered a practicable approach for patients with similar conditions.

In addition to hydrosalpinx caused by inflammation, our report proved the presence of hydrosalpinx resulting from the interruption from ampulla and fimbria, which is associated with congenital fallopian tube development. None of these patients exhibited signs of pelvic infection or adhesions during laparoscopy; therefore, we considered their hydrosalpinx not to be due to infection. During the sixth week of female embryonic development, the epithelium of the lateral body cavity at the head end of the mesonephric duct depresses into the mesenchyme, forming a longitudinal sulcus that develops caudally toward the embryo. Subsequently, the edges of the sulcus merge to form a duct on each side called the Müllerian duct. The cranial portion of these ducts communicates with the abdominal cavity and forms what is referred to as the tubal section. The fallopian tube's cranial end remains open with multiple invaginations along the free edge, which developed into the fimbriae.[1] Any abnormalities occurring during any stage or part of this developmental process can lead to anomalies in Müllerian system development.

Classification systems of Müllerian malformations have undergone successive modifications.[5] In reality, malformations within an organ system do not adhere to a particular pattern. Abnormalities affecting Müllerian systems may involve various organs, such as the uterus, cervix, vagina, and tubes. Congenital anomalies related to fallopian tubes include aplasia, hypoplasia, accessory ostia, paratubal cysts, accessory tubes, and congenital diverticula.[6] Case 1 exhibited bilateral interruption of the ampulla and the fimbria; therefore, according to the Vagina, Cervix, Uterus, Adnexa, and Associated Malformations classification system,[7] this case would be classified as follows: normal vagina,

TABLE
Summary of congenital interruption between the ampulla and fimbria cases

Case	Age, y	Infertility, y	Infertility type	Occlusion site	Other malformations	Therapies	Method of conception	Fertility outcome
1	30	2	Tubal factor	Bilateral	None	Bilateral fallopian tube ampullastomy	Spontaneous	25 wk of pregnancy
SP1	32	4	Male factor	Left	None	Left tubal ligation	Spontaneous	1 girl in 2019
SP2	31	1	Tubal factor	Left	None	Left fallopian tube ampullastomy	/	/
SP3	29	2	Tubal factor	Left	None	Left fallopian tube ampullastomy	/	/
SP4	34	6	Tubal factor	Right	None	Right tubal ligation	/	/
SP5[a]	29	2	Tubal factor	Left	None	Left salpingectomy	/	/

ART, assisted reproductive technology; *SP*, supplementary.

[a] This case is from Dahan et al.[8]

Dong. Congenital interruption between ampulla and fimbria of the fallopian tube. Am J Obstet Gynecol Glob Rep 2024.

cervix, and uterus; bilateral tubal malformation, ovaries normal; and no associated malformation. In contrast, the supplementary cases were all unilateral and classified as follows: normal vagina, cervix, and uterus; unilateral tubal malformation, ovaries normal; and no associated malformation.

The partial absence of the fallopian tube is usually associated with major ipsilateral uterine malformations, including bicornuate uterus and ipsilateral nonfunctional uterine horns.[8] Minor tubal malformations can be easily neglected, particularly in women undergoing assisted reproductive treatment. In this article, we discuss 5 similar tubal malformations characterized by incomplete continuity between the ampulla and the fimbria; the ampulla is blind-ended, which leads to hydrosalpinx and may be a significant cause of infertility. This resembles the case[8] previously reported, but in this case, there was no subsequent pregnancy outcome, and salpingectomy was performed as the only option.

Tubal malformations constitute a minority of anomalies in the female reproductive tract. The potential mechanism involves either segmental torsion with reabsorption or a vascular accident.[8] A study[9] investigating the origin of tubal epithelial cells revealed that the distal and proximal populations of the fallopian tubes are distinct lineages unique to early tubal development and are maintained separately, suggesting a possible temporal or spatial disparity in the development of proximal and distal cells leading to disruption of the ampulla, which requires further confirmation.

Although Müllerian duct malformations may be associated with urinary malformations,[10] no significant malformations except for the fallopian tubes were observed in the cases included in this study. In addition, 2 of these cases had already conceived or given birth after surgery, suggesting that this phenomenon may not have a substantial impact on uterine function and does not seem to be correlated with renal malformations. It may be related to early developmental abnormalities during the embryonic period.

Conclusion

In conclusion, in patients with infertility, discontinuity between the ampulla and the fimbria can lead to hydrosalpinx, which affects conception. For such patients, hydrosalpinx can be treated by salpingostomy, after which natural conception can be tried, or assisted reproductive treatment can be performed after salpingectomy. ■

CRediT authorship contribution statement

Xiaotong Dong: Writing – original draft. **Shengrui Zhao:** Writing – review & editing. **Peng Liu:** Data curation. **Qian Yu:** Data curation. **Lei Yan:** Visualization, Resources.

ACKNOWLEDGMENTS

We thank all the physicians and scientists for their assistance with data collection as well the patients for participating in this study.

Supplementary materials

Supplementary material associated with this article can be found in the online version at doi:10.1016/j.xagr.2024.100353.

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