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Short Communication

Congenital Tubal Anomalies: Exploring Their Relationship with Tubal Factor Infertility an Observational-Descriptive Study

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Abstract

Background: Infertility represents a significant global concern affecting millions of couples worldwide. Among the various causes contributing to infertility, tubal factor infertility (TFI) emerges as a prominent factor that warrants closer investigation. Such abnormalities may originate from multiple causes, including congenital tubal anomalies (CTAs). This study aimed to reveal the prevalence of CTAs and associated pathologies among patients diagnosed with TFI. **Methods**: This observational study was conducted at two centers (University Hospital and a referral infertility clinic), between 2020 and 2023. Diagnostic laparoscopy was offered to all women diagnosed with TFI after hysterosalpingography (HSG) and who did not achieve pregnancy within six months after the examination. Patients who consented to undergo diagnostic laparoscopic were included in the study. **Results**: After the initial infertility work-up, 895 women (21%) were diagnosed with suspected TFI. Out of these, 220 women consented to undergo diagnostic laparoscopy. Among them, 113 (51.3%) were found to have surgically confirmed tubal pathologies/anomalies, while 107 (49.7%) revealed normal findings. A total of 34 women (15%) were found to have, total or partial agenesis, among which 32 revealed congenital uterine anomalies, and 16 revealed other system anomalies. **Conclusions**: CTAs contribute significantly more to TFI than expected in the general population. Therefore, careful systemic examinations, especially for uterine anomalies, are essential.

Keywords: congenital tubal anomalies (CTAs); hysterosalpingography (HSG); tubal factor infertility (TFI)

1. Introduction

The fallopian tubes play a crucial role in the reproductive process. These structures typically measure 10–12 cm in length and are comprised of four parts from the proximal to the distal side: intramural, isthmic, ampullary, and fimbria [1]. Any abnormalities in their development or structure can severely affect fertility. One such anomaly is fallopian tube agenesis, whether partial or complete. Partial fallopian tube agenesis refers to the absence or shortening of a segment of one or both tubes, while complete agenesis indicates the complete absence of the fallopian tubes. These anomalies are frequently reported in conjunction with uterine and other system malformations [2].

General tubal factor infertility (TFI) is very common, defined as either blocked fallopian tubes or inability of the tubes to pick up an oocyte from the ovary, accounts up to 67% of infertility diagnoses, depending on the population studied [3]. On the other hand, congenital tubal anomalies (CTAs) are very rarely reported in the literature, mostly case series, including agenesis, hypoplasia, accessory ostia, paratubal cysts, accessory tube, accessory ampulla, multiple luminal, unilateral absence of a fallopian tube, total or partial duplication of fallopian tube, tubal dislocation, and complete and segmental absence of a portion of the fallopian tubes [4,5]. Up to date, not many data have evaluated the relation between TFI and CTAs and this relation remains an enigmatic entity.

The aim of this study is to review a case series of partial or complete fallopian tube agenesis, whether unilateral or bilateral, by examining its prevalence and reviewing associated anomalies in cases of primary infertility cases.

2. Material and Methods

This observational study was conducted at two centers (University Hospital and a referral infertility clinic), between 2020 and 2023. All couples presenting with primary infertility were evaluated in an outpatient clinic using fundamental hormone analysis, semen analysis, sonography, and hysterosalpingography (HSG) as part of the standard of care. Women aged between 18 and 40 years, without confirmed chromosomal anomalies, were included in the analysis. In suspected cases, 3D vaginal sonography was performed to clarify any uterine anomalies. Laparoscopy (L/S) was offered to patients who could not conceive within six months after the initial examination, and those who accepted were included in the study. All women with suspected tubal pathology identified during HSG were offered diagnostic laparoscopy to confirm the anomaly. After obtaining informed consent, two experienced surgeons performed the surgeries. When tubal and Müllerian anomalies were detected, examination of the urinary system was conducted using intravenous pyelography. Other suspected pathologies were examined by abdomen-pelvic computed tomography (CT) X-ray imaging. All

anomalies were reported in an electronic database. Exclusion criteria were as follows: (1) women without laparoscopically confirmed tubal anomalies; (2) incomplete infertility follow-up; (3) incomplete anomaly screening; (4) incomplete medical records; (5) cases of secondary infertility, and (6) women with a history of prior tube surgery. Prior to participation in the study, all subjects provided informed consent for inclusion. The study protocol was approved by the Ethics Committee (approval number 2020-0001/20) and conducted in accordance with the Helsinki Declaration.

3. Results

During the study period, a total of 4230 infertile couples were admitted to the clinics. The mean age of women in the study population is 30 $\pm$ 2.3 years. The overall duration of infertility is 1.2 $\pm$ 0.6 years. Among them, 895 women were diagnosed with suspected tubal pathology following the HSG examination (No = 895/4230) (Overall prevalence 21%). Baseline demographic characteristics, such as age, body mass index (BMI), duration of infertility, and sperm concentration, are shown in Table 1. Diagnostic L/S was performed on 220 women. A flowchart of the patients is given in Fig. 1. Among them, 113 (51.3%) women were found to have tubal pathologies/anomalies, while 107 (48.6%) showed normal findings. A total of 34 women revealed congenital proximal and distal tubal anomalies. Half of these cases (17 out of 34) involved complete agenesis, while the remaining cases presented with proximal or mid-distal portion agenesis. The final clinical and pathologic findings are given in Table 2. The majority of women, comprising 32 out of 34 (94%) with tubal pathology, also demonstrated at least one uterine anomaly. Among them, 19 (55.9%) patients were confirmed to have a unicornuate uterus, 6 (17.6%) had uterus didelphys, 4 (11.8%) had a bicornuate uterus, and 3 (8.8%) had a uterine septum. No uterine anomaly was observed in 2 (5.9%) patients. A total of 16 women were diagnosed with other systemic anomalies, including those affecting the urinary tract, gastrointestinal, or musculoskeletal. These findings are summarized in Table 3.

Table 1. Baseline characteristics of the patients (N = 113).

Age, years	30 $\pm$ 2.3
BMI, kg/m^2	24.7 $\pm$ 4.31
Duration of infertility, years	1.2 $\pm$ 0.6
Sperm concentration/mL	22.8 $\times$ 10^6 $\pm$ 3.6 $\times$ 10^6

BMI, body mass index; N, number.

4. Discussion

This observational study showed that TFI is one of the major etiological factors among primary infertility cases,

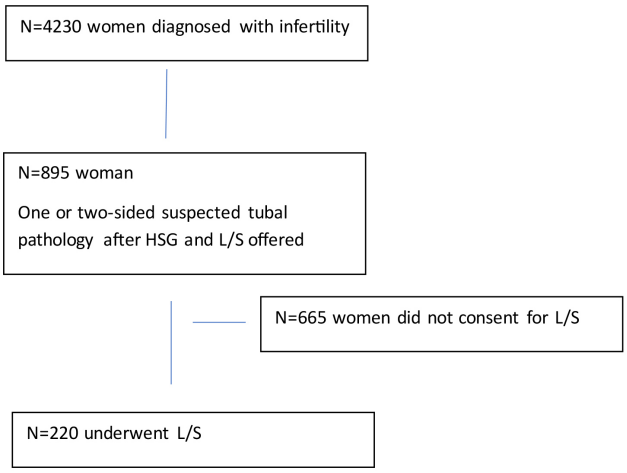

Fig. 1. Flowchart of the patients after initial infertility investigation. HSG, hysterosalpingography; L/S, Laparoscopy.

and CTAs were reported to be relatively higher (15%) in younger women, contrary to general expectations.

Partial or complete absence of a fallopian tube is very rarely reported in the literature [4,6]. Even the congenital absence of a single fallopian tube segment has been reported [6]. Hence, the first topic to be addressed should be the high incidence of CTAs in our series. Clarification of the incidence estimation in the general population is necessary as no data has been published in this context. The exact incidence of tubal agenesis, exceptionally one-sided, may be higher than reported, as the morphologically and functionally regular contralateral tubes can lead to spontaneous pregnancies. Thus, it remains unclear whether the unilateral absence of the tube and ovary is a contributing factor of infertility. We recommend a thorough laparoscopy examination of both fallopian tubes after having suspected HSG results, especially in young patients presenting with primary infertility cases.

Congenital tube anomalies encompass either the complete agenesis of an entire tube or segmental agenesis, which may involve the proximal-mid or distal segments. In some cases, ipsilateral agenesis of an ovary is likely to accompany tubal agenesis [4]. The congenital absence of the ovary and fallopian tube is rare, and there is insufficient information about this anomaly in the literature [7,8]. Most patients are asymptomatic and may be diagnosed incidentally during intraabdominal exploration or laparoscopy. The actual etiologies of ipsilateral tubal and ovarian absence remain unclear. Three possible etiologies may be responsible: (I) adnexal torsion, (II) tubal and ovarian maldevelopment secondary to ischemia due to a vascular accident, and (III) a defect in the development of the Müllerian and mesonephric system, either entirely on one side or localized to the region of the genital ridge and the caudal part [4].

A second important finding is the high incidence of congenital uterine anomalies in women with CTAs. In our

Table 2. Pathologic laparoscopic findings of patients (N = 113).

Congenital tubal anomalies (CTA) 34 (15%)		Acquired tubal pathologies 79 (35.9%)	
Unilateral complete agensis	18	Phimosis (unilateral)	11
Unilateral proximal-isthmic agenesis	3	Proximal tubal occlusion	28
Unilateral proximal and mid-portion agenesis	3	Unilateral Hydrosalpinx	24
Unilateral distal and mid-portion agenesis	7	Bilateral Hydrosalpinx	14
Unilateral distal agensis	3	Isthmica nodosa	2

CTA, congenital tubal anomalies; N, number.

Table 3. Uterine, genital, and other system anomalies detected in women with CTA (N = 61).

Congenital uterine anomalies (N = 32)		Congenital lower genital tract anomalies (N = 13)		Other system anomalies (N = 16)	
Unicornuate	19	Unilateral absence of the ovary	6	Urinary system anomaly	10
Septate	6	Unilateral rudimentary ovary	6	Accessory spleen	3
Bicornuate	4	Vaginal and cervical hypoplasia	1	Agenesis of the vermiform appendix	1
Didelphys	3			İpsilateral ureteral duplication	1
				Musculoskeletal	1

CTA, congenital tubal anomalies; N, number.

series, the majority of women with CTA revealed uterine anomaly (32/34), mostly unicornuate uterus. Septate, unicornuate, and bicornuate uterus are frequently associated with tubal agenesis [2]. A recent review, which includes several case reports, has documented incidental findings of bilateral or unilateral agenesis of the tubes and uterine malformations [9]. These findings have been confirmed by others [10]. These concomitant anomalies may result from shared embryological origins or abnormal developmental processes during early fetal life, such as a defect in Müllerian duct development or, more likely, to a defect in the genital ridge region. The exact nature of such anomalies is still not entirely elucidated and needs further investigation. However, clinicians should prioritize the evaluation of the uterus when sign of tubal pathology are detected during examinations, particularly in cases with congenital tubal anomalies. This is critical for counselling patients about the risks associated with future pregnancy.

The third important point is the discordant findings between pathologic HSG and L/S. Nearly half of the women with unilateral or bilateral tubal pathology revealed normal L/S findings. This is consistent with recent data, as studies have reported that the reliability of HSG in diagnosing tubal occlusion and patency is questionable, particularly due to lower genital tract spasms [11–14]. According to sensitivity analysis, HSG is reported to have limited sensitivity to detect tubal pathologies [13,14]. Therefore, it is advisable to consider L/S for evaluating tubal patency before initiating advanced fertility treatments such as *in vitro* fertilization (IVF). If CTA is detected, it is essential to perform a careful examination of the uterine cavity using 3D sonography, especially for young age women who desire fertility.

Finally, CTA are frequently associated with additional system anomalies, particularly urinary tract anomalies. Common pathologies associated with congenital tubal anomalies include unilateral renal agenesis, horseshoe kidneys, pelvic kidney, and ipsilateral double ureter. The incidence of unilateral renal agenesis is 1 per 500–1000 autopsies and 1 per 2900–3200 births [15]. Close embryological development may result in the association of unilateral renal agenesis with other mesonephric and Müllerian ductal anomalies. In our study, urinary system anomalies were detected in 10 out of 34 CTA cases (29%) of patients with tubal anomalies. This underscores the importance of clinicians being aware of the presence of severe urinary tract anomalies in such women, even at a young age.

A significant limitation of our descriptive is the absence of a control group from an average population to compare incidences. A larger sample is required to draw more precise conclusions about the exact provinces precisely. We did not report fertility data following L/S. On the other hand, all of our cases were laparoscopically confirmed anomalies, and all procedures were performed by the same surgeons. Moreover, only a minority of women consented to diagnostic L/S, given its surgical nature. More descriptive studies with a larger sample size are needed.

5. Conclusions

CTAs may represent an unexpected cause of tubal factor infertility underscoring the importance of carefully reviewing other system anomalies, especially uterine anomalies.

Availability of Data and Materials

All data points generated or analyzed during this study are included in this article and there are no further underlying data necessary to reproduce the results.

Author Contributions

EP and RP designed the research study. Gİ and DD performed the research. All authors contributed to editorial

changes in the manuscript. All authors read and approved the final manuscript. All authors have participated sufficiently in the work and agreed to be accountable for all aspects of the work.

Ethics Approval and Consent to Participate

All subjects gave informed consent for inclusion before participating in the study. The study was conducted in accordance with the Declaration of Helsinki, and the protocol was approved by the Ethics Committee of the Centrum Ankara Womens Health and IVF Center in affiliation with Ufuk University School of Medicine (approval number 2020-0001/20).

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Conflict of Interest

The authors declare no conflict of interest.

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