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Case Report

A rare case of primary infertility with bilateral agenesis of medial part of fimbrial end with hypoplasia of fimbria and absence of fimbria ovarica with septate uterus with bilateral normal ovaries

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ABSTRACT

Patients with infertility frequently present with associated congenital genital anomalies affecting around 5-6% of patients. Most of these patients have anomalies associated with uterus, cervix and vagina which have been extensively studied through time. The exact association of abnormalities of fallopian tube to infertility is still unknown due to the limited data available. The true incidence of congenital fallopian tube anomalies is unknown because abnormalities may be subtle and are often overlooked or thought to be due to acquired or iatrogenic causes. While reviewing the literature, we observed that partial or complete agenesis of the fallopian tube is rarely reported. They are usually incidental diagnosis on laparoscopy done for some other purposes. Due to rarity of such cases, their effect on fertility and its management is still a challenge. Here, we present a case report of septate uterus with bilateral fimbrial agenesis and normal ovaries in a patient of primary infertility.

Keywords: Fimbrial agenesis, Infertility, Congenital anomalies, Laproscopy

INTRODUCTION

Patients with infertility frequently present with associated congenital genital anomalies affecting around 5-6% of patients.[1] Most of these patients have anomalies associated with uterus, cervix and vagina which have been extensively studied through time. The exact association of abnormalities of fallopian tube to infertility is still unknown due to the limited data available.

Fallopian tube development is thought to take place during embryonic differentiation of the paramesonephric or "Müllerian" ducts. In the sixth week of gestation, the bilateral Mullerian ducts migrate towards the midline and fuse to form the uterus and upper one-fifth of the vagina. Rostrally, the Mullerian ducts form fallopian tubes. Any disturbance in the migration, fusion or resorption of these ducts may result in Mullerian anomaly.[1]

There are few proposed theories for etiopathogenesis of bilateral adnexal malformations.[2] The first theory includes asymptomatic adnexal torsion during childhood or adulthood. This is followed by auto-amputation and resorption of the affected segment. The other proposed theory is congenital agenesis of distal part of Mullerian duct or a defect in localised region of genital ridge. In comparison to Mullerian duct-derived organs, congenital defects of the ovary are rare. Gonadal development depends on germ cell migration, as well as appropriate formation of the urogenital ridge. These processes are regulated by multiple factors and genes, and a unilateral defect at any point during this process may prevent ovarian formation. It has been hypothesized that a defect localized to the region of the genital ridge and the caudal area of the Mullerian duct reflects improper development of the urogenital ridge, which affects the development of the fallopian tube in that region. Several studies have indicated that an inadequate blood supply during the descent into the

pelvis of the caudal section of the paramesonephric duct may lead to adnexal agenesis. However, a clear developmental explanation for this malformation has not yet been elucidated.

The true incidence of congenital fallopian tube anomalies is unknown because abnormalities may be subtle and are often overlooked or thought to be due to acquired or iatrogenic causes.[2]

While reviewing the literature, we observed that partial or complete agenesis of the fallopian tube is rarely reported. They are usually incidental diagnosis on laparoscopy done for some other purposes.[2] Due to rarity of such cases, their effect on fertility and its management is still a challenge.

Here, we present a case report of septate uterus with bilateral fimbrial agenesis and normal ovaries in a patient of primary infertility.

CASE REPORT

Mrs. J, 27-year-old female came to our IVF OPD as a case of primary infertility. Patient was married for 4 years and cohabiting with her husband regularly. She had menarche at the age of 14 years and was having regular menstrual cycles with normal flow since then. Her father was diabetic and there was no other significant past or family history. She had undergone 3 cycles of ovulation induction previously at some other centre and her HSG was wrongly reported as bicornuate uterus with bilateral distal tubal blockage (Figure 1). Other investigations including husband semen analysis, antral follicle count and hormone profile were normal.

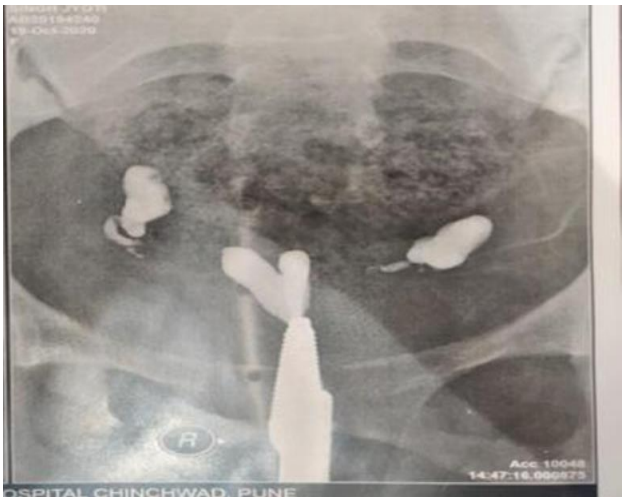

Figure 1: HSG showing septate uterus wrongly reported as bicornuate uterus with bilateral distal tubal blockage.

However, we performed her 3D ultrasonography which showed the presence of complete uterine septum (Class U2bC0V0) along with bilateral hydrosalpinx. There were no associated renal or gastrointestinal anomalies. She was taken up for operative laparoscopy and hysteroscopy. Her consent for SOS bilateral salpingectomy was taken and further need for IVF was explained to the patient and her husband.

Hysteroscopy showed cavity with complete uterine septum. Bilateral ostia were visualised as normal. Complete resection of septum was done hysteroscopically with scissors (Figure 2).

Laparoscopy showed bilateral hydrosalpinx with bilateral agenesis of medial part of fimbrial end with hypoplasia of fimbria and absence of fimbria ovarica (Figure 3 and 4). Chromopertubation test showed absence of bilateral spillage of dye confirming our visual findings. Bilateral ovaries and external contour of uterus was visualised as normal. On both the sides, single ureter was visualised as normal without dissection of the lateral pelvic wall (Figure 5). Bilateral salpingectomy was done as close to fallopian tube as possible (Figure 6) without compromising the ovarian blood supply. Specimen was sent for histopathology (Figure 7) which confirmed our findings. The rest of the abdomen appeared normal and there were no signs of Koch's abdomen, endometriosis or pelvic infection.

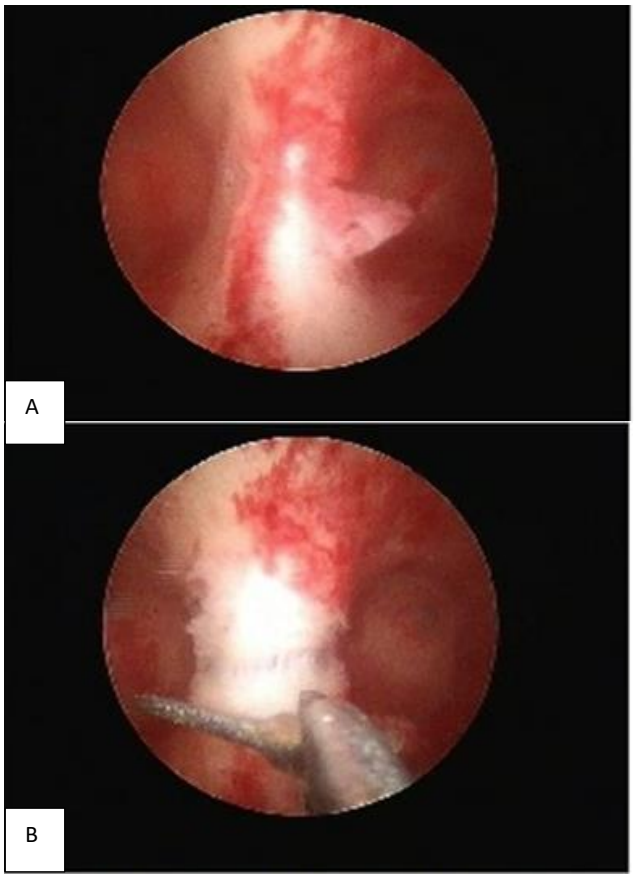

Figure 2 (A and B): Hysteroscopy showing complete uterine septum and septal resection in progress using scissors shown.

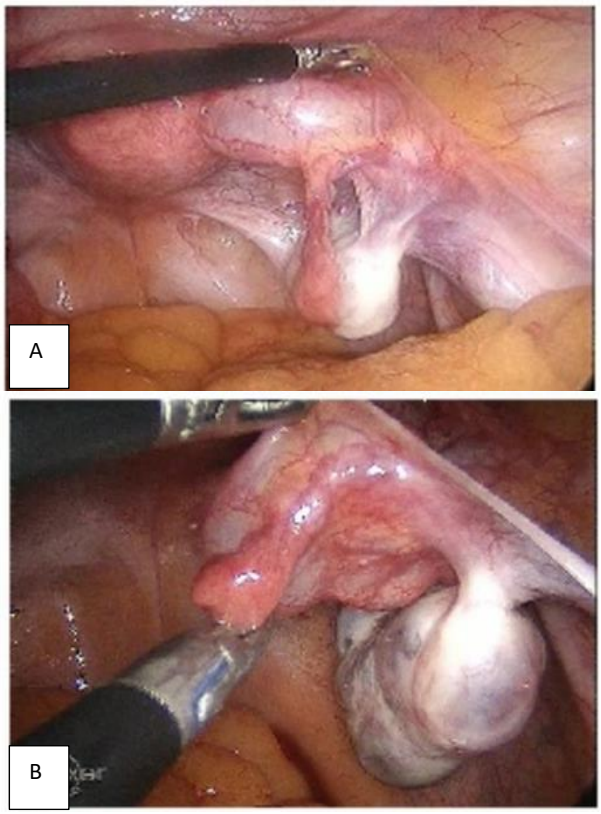

Figure 3 (A and B): Right sided hydrosalpinx with hypoplastic fimbria and agenesis of medial portion of fimbrial end and absence of fimbria ovarica.

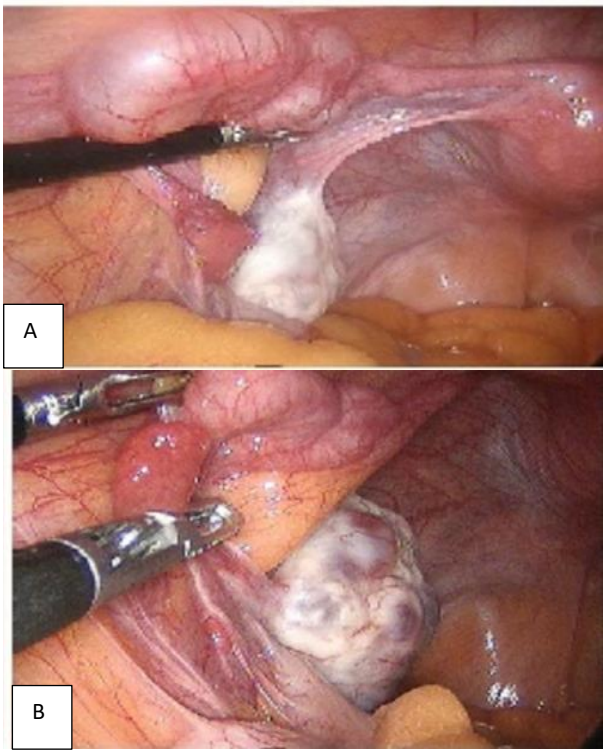

Figure 4 (A and B): Left sided hydrosalpinx with hypoplastic fimbria and agenesis of medial portion of fimbrial end and absence of fimbria ovarica.

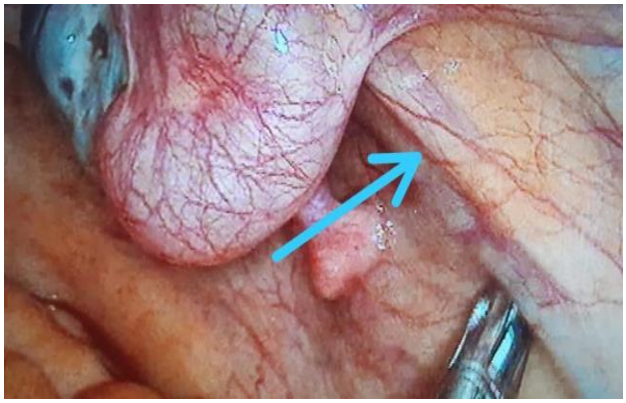

Figure 5. Normal single ureter visualized on the lateral pelvic wall.

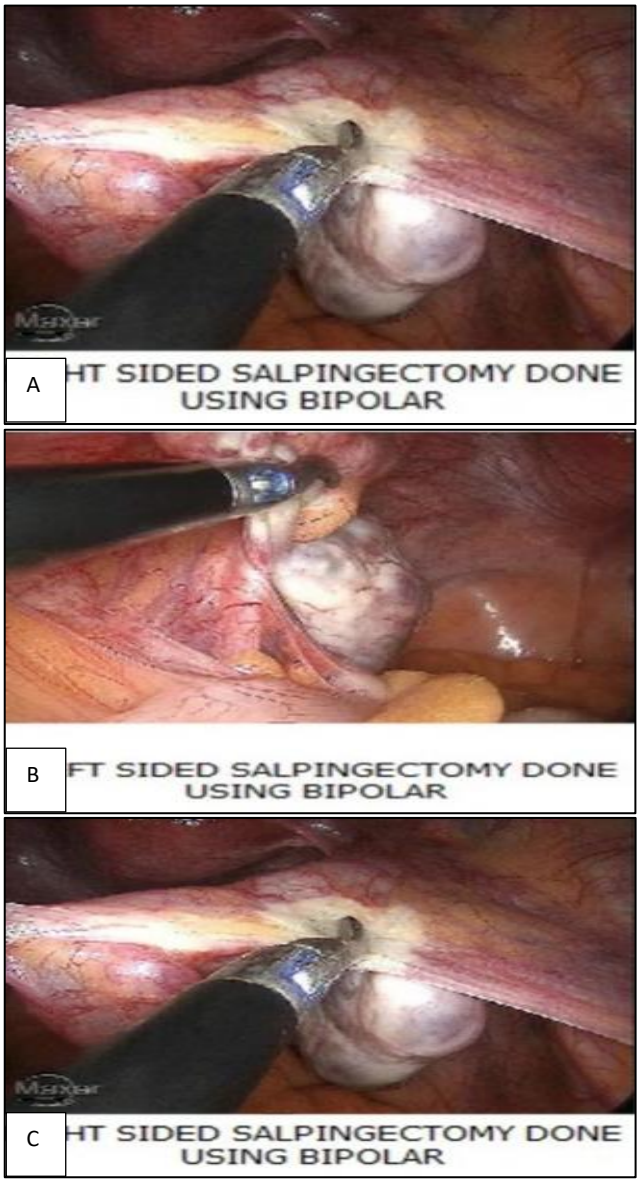

Figure 6 (A-C): Bilateral salpingectomy done as close to fallopian tube as possible without compromising the ovarian blood supply. End result showing normal uterus and ovaries.

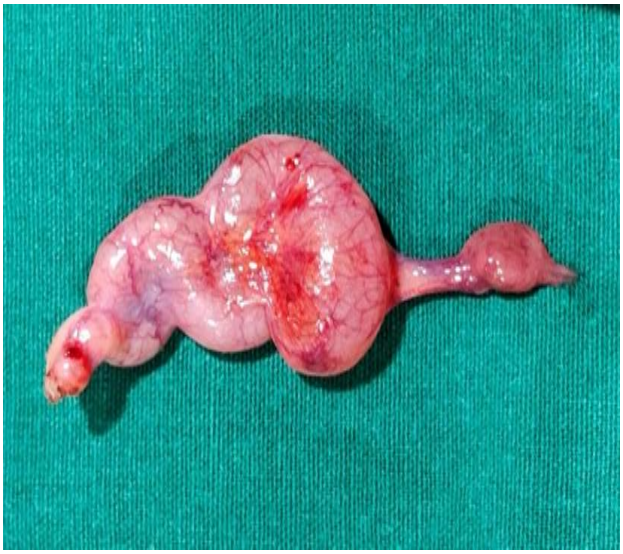

Figure 7: Specimen of hydrosalpinx with hypoplastic fimbria and agenesis of medial portion of fimbrial end.

DISCUSSION

Review of literature shows very rare occurrence of congenital fallopian tube anomalies.[2] These patients are usually asymptomatic or diagnosed incidentally on ultrasound or laparoscopy done for infertility. Due to rarity of cases, there are no accepted standard classifications available for such anomalies. The CONUTA (Congenital uterine anomalies) classification which is used for Mullerian anomalies does not give a standard classification for fallopian tube abnormalities.[3] VCUAM (Vagina cervix uterus adnexaa-associated malformation) classification was proposed. This classification also includes tubal and ovarian abnormalities. Our case falls in category A1b of the VCUAM classification.[4] There proposed theories for etiopathogenesis of bilateral adnexal malformations are asymptomatic adnexal torsion during childhood or adulthood and congenital agenesis of distal part of Mullerian duct.[2]

A few similar cases have been reported in the past. Nawroth et al reviewed cases of 18 patients with partial atresia of fallopian tube.[5] Interestingly, 14 out of the 18 patients had unilateral tubal atresia while only 4 had bilateral involvement. But the combination of partial atresia of tube along with bicornuate uterus in 5 cases pointed to congenital origin of the anomaly.

Yucuku et al reported a case series of 4 cases of unilateral partial tubal agenesis.[6] Three out of the 4 cases had ovarian agenesis of the same side whereas one case showed adnexal torsion of the opposite side. There was no association of other congenital anomalies of Mullerian structures. They concluded that accidental adnexal torsion in childhood can explain such cases.

Chen et al reported a similar case of unilateral ovarian and Fallopian tube agenesis in a patient of infertility with normal uterus.[2] They supported the torsion hypothesis as there was no associated uterine anomaly.

Shrotri et al reported a case of bilateral absence of fallopian tube.[7] In the absence of any previous infection or torsion and due to normal uterus and ovaries, they concluded that the aetiology was likely congenital.

Our case is one of the rare cases with congenital complete uterine septum and bilateral agenesis of medial part of fimbrial end with hypoplasia of fimbria and absence of fimbria ovarica.

As there was no history of previous surgery and infection and in the presence of associated uterine septum with confirmatory laparoscopy and histopathology findings, diagnosis of a congenital bilateral agenesis of medial part of fimbria end with hypoplasia of fimbria and absence of fimbria ovarica is more likely.

CONCLUSION

Our case is a rare case of congenital bilateral agenesis of medial part of fimbrial end with hypoplasia of fimbria and absence of fimbria ovarica and it remains asymptomatic till fertility is desired.

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