

Does the fimbria have an embryologic origin distinct from that of the rest of the fallopian tube?

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Objective: To propose a new theory describing the development of the fallopian tube fimbria.

Design: Case series report.

Setting: Metropolitan tertiary care children's hospital.

Patient(s): Two girls, aged 12 and 20 years, who presented with pelvic pain.

Intervention(s): Magnetic resonance imaging, laparoscopy with salpingectomy, and pathologic analysis.

Main Outcome Measure(s): Description of a novel theory regarding the embryologic development of the fallopian tube and its fimbria.

Result(s): In two non-sexually active girls the cause of their pelvic pain was found to be a hydrosalpinx associated with a discontinuous fallopian tube in which the fimbriated end did not directly communicate with the remainder of the fallopian tube.

Conclusion(s): The two cases of pure congenital fallopian tube atresia, the presence of fimbriae in patients with müllerian (uterine, cervical, and vaginal) agenesis, and the role of the fimbria in ovarian-like and peritoneal cancers, support a novel hypothesis that the fimbria of the fallopian tube may arise separately from the rest of the tube. (Fertil Steril® 2008;90:2008.e5–e8. ©2008 by American Society for Reproductive Medicine.)

Key Words: Fallopian tube, müllerian anomaly, fimbria, congenital fallopian tube atresia, embryology

The fallopian tube is believed to be of müllerian derivation. Fallopian tube formation is thought to take place during embryonic differentiation of the paramesonephric or "müllerian" ducts.

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 (1). It is estimated that up to 70% of tubal malformations occur in the context of associated renal and/or uterine anomalies (2). As with many congenital anomalies, the pathogenesis is often traced back to embryologic development. Most müllerian anomalies can therefore be explained by a failure or dysregulation of events that occur during embryologic development. Some but not all congenital malformations of the fallopian tube can be explained as disruptions of the currently accepted sequence of normal development. Cases of "congenital" interruptions or occlusions of the fallopian tube have been reported; these

reports are often confounded by patient history of infection or trauma (3, 4).

We report two cases of discontinuous fallopian tubes in non-sexually active adolescent girls in which there was absence of a lumen and a muscular wall between the fimbriated end and a blind-ending proximal portion of fallopian tube. We discuss these findings, in particular with respect to developmental anomalies in müllerian agenesis as well as the hypothesis of fimbrial involvement in ovarian-like carcinogenesis.

CASE REPORTS

Case One

An 11-year-old girl was referred to the Gynecology Program at Children's Hospital Boston for evaluation of abdominal pain and a pelvic mass. A pelvic ultrasound revealed a 5-cm cystic structure in the left adnexa, likely representing a dilated fallopian tube. Follow-up magnetic resonance imaging confirmed this diagnosis and was otherwise unremarkable, demonstrating a normal uterus, right tube and ovary, and normal kidneys. Her medical history was noncontributory. She had no prior surgeries. Menarche occurred at age 9, and she reported regular periods without significant dysmenorrhea. She had no history of sexual activity or of

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sexual abuse. An operative laparoscopy was performed and revealed a left hydrosalpinx. The fimbria appeared grossly normal without evidence of scarring or adhesions. There did not seem to be direct communication between the fimbriated end of the tube and the remainder of the tube itself. Chromopertubation resulted in dilation of the hydrosalpinx with blue dye but no spillage from the fimbriated end. Because there was no normal fallopian tube lumen attached to the fimbriated end of the tube, reanastomosis was not possible and a salpingectomy was performed. Microscopically, the dilated proximal portion of fallopian tube showed simplified plicae and mural fibroplasia with scattered eosinophils and mast cells (Fig. 1). The fimbriated end showed well-developed fimbria with no specific pathologic features.

Case Two

A 20-year-old nulligravid woman was referred for chronic pelvic pain, vaginal discharge, and a right hydrosalpinx found incidentally on computed tomographic scan for pyelonephritis 3 years earlier. There were no known sequelae of this uncomplicated infection; however, given the persistence of cyclic pelvic pain she was referred to the Gynecology Program for evaluation. Her gynecologic history included menarche at age 14 with regular menses and an imperforate hymen (surgically repaired at age 17 years). She complained of chronic cyclic pelvic pain that was not relieved by ovulation suppression or nonsteroidal medications. She was never sexually active and denied any history of pelvic infections, sexually transmitted diseases, or sexual abuse. On physical examination, she had Tanner stage 5 breast development with normal-appearing external genitalia. Cervical cultures for gonorrhea and chlamydia were negative. Subsequent

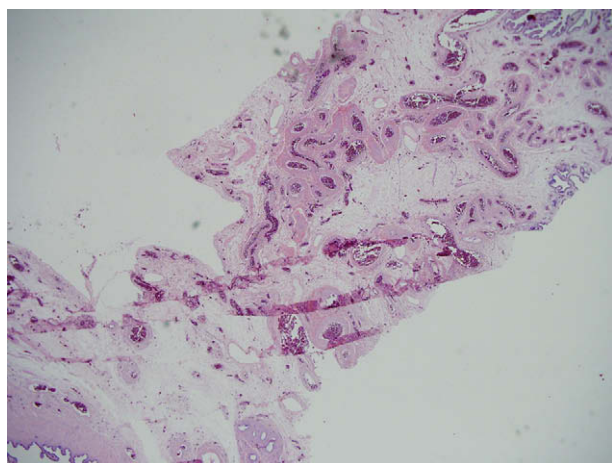
magnetic resonance imaging revealed complete resolution of the hydrosalpinx with normal anatomy, including normal kidneys, uterus, and adnexa bilaterally. She continued to experience pain despite negative imaging and therefore underwent diagnostic laparoscopy. Surgical exploration showed a right hydrosalpinx. There was no evidence of inflammation, endometriosis, or adhesions. Again, the tube was felt to end blindly with no direct communication to the fimbriated end. The fimbriae appeared normal except for their lack of connection to the blind-ending proximal portion of the tube. A right salpingectomy was performed. Gross pathologic examination confirmed the intraoperative findings. Discontinuity of the tube was noted, with a blind-ending proximal portion of the tube dilated with a hydrosalpinx, and a separate fimbriated end. The two segments were connected only by a thin web of mesothelium-lined fibroconnective tissue (Fig. 2). Microscopic examination confirmed the lack of epithelial and muscular continuity. The proximal fallopian tube showed a well-developed muscular wall and a simplification of the plical architecture, as is typical of hydrosalpinx. The fimbriated end showed robust fimbriae with no pathologic features. There was no fibrosis, hemosiderosis, or chronic inflammation. Her pain was perhaps explained by waxing and waning of the hydrosalpinx with reabsorption of the hydrosalpinx and subsequent reaccumulation. There was no evidence grossly or microscopically of intermittent torsion, scarring, or prior infection.

DISCUSSION

Anomalies of the fallopian tube, including complete absence, partial absence, duplication, and ectopic location,

FIGURE 1

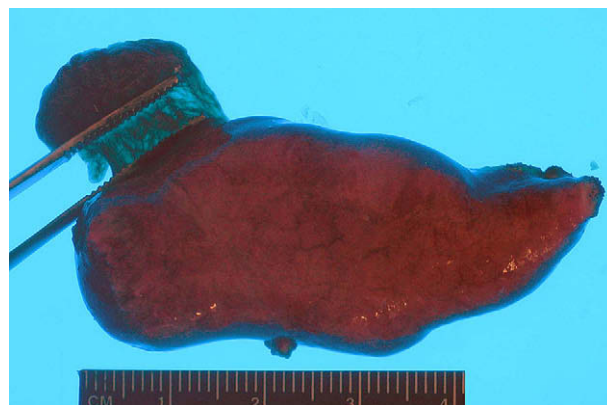
Microscopic examination showed a cord of vascularized fibrous tissue connecting the proximal fallopian tube (lower left) and the fimbria (upper right) (original magnification, $\times 2$).



Garrett. Development of fallopian tube fimbria. *Fertil Steril* 2008.

FIGURE 2

The resected specimen included a dilated segment of proximal fallopian tube and a fimbriated end (upper left), connected only by a thin fibrous membrane without any muscular wall. Transillumination shows a fine vascular network with vessels of a similar caliber and density to those of the pelvic peritoneum and its reflections.



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have all been described (10). Partial tubal atresia has been described both with and without uterine anomalies. When associated with a uterine malformation, it is typically seen with bicornuate uteri, whereby the tube ipsilateral to the rudimentary, nonfunctioning horn contains an atretic ampullary segment (5). In these cases the fimbriated end is connected to a shortened, patent ampullary segment that ends blindly in the mesosalpinx. Polasek et al. (6) reported bilateral tubal atresia associated with a transverse vaginal septum; in this patient, the mid-portions of both fallopian tubes were absent, and the authors did not speculate about the etiology.

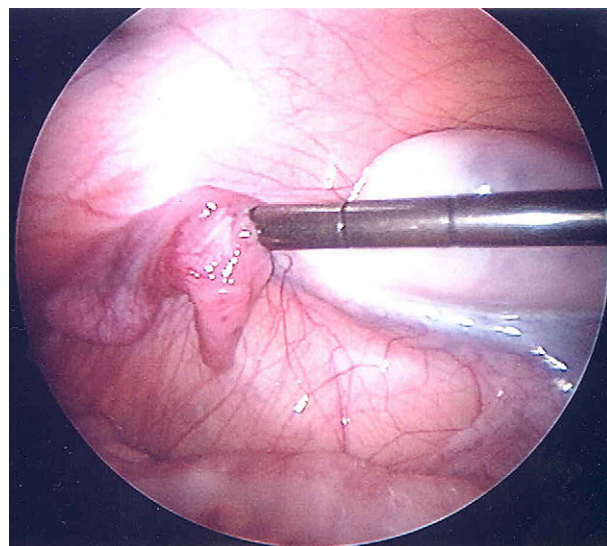
Seven cases grossly similar to the patients reported herein have been described and termed *congenital ampullary atresia* (3, 7). The histologic features in these cases were not described in detail. Investigators have speculated that the atresia arises from failure of formation of the ampulla's distal segment, leaving an obstructed mid-ampullary segment connected to isolated fimbrial tissue through a web of mesosalpinx, probably resulting from torsion or other vascular accident (7). Our investigation included thorough histologic examination in both cases. Submission of all tissue from both cases failed to identify any microscopic evidence of hemosiderosis, a finding that is typical in adnexal torsion due to the profound venous congestion that precedes infarction. Although we can not absolutely rule out torsion in the two patients presented herein, their clinical histories and absence of histologic sequelae strongly contest such a theory.

A well-known cause of acquired tubal atresia is infection. In this respect it is notable that the two adolescent patients reported herein were not sexually active, had no history of sexual abuse, and clearly had no evidence of previous infection. This is in contrast to the five previously reported patients with similar gross findings, all of whom were older and sexually active, and three of whom had a history of chlamydia infection. By demonstrating a similar anomaly in young, non-sexually active girls, we support the hypothesis that true congenital ampullary atresia is a defined entity separate from that of acquired tubal occlusion.

Müllerian anomalies encompass a spectrum of heterogeneous malformations demonstrating the complexity of timing and regulation of embryologic events that can go awry. Pure müllerian agenesis, or Mayer-Rokitansky-Küster-Hauser (MRKH) syndrome, consists of a complex malformation in which the vagina is absent and the uterus and fallopian tubes are either absent or rudimentary. The findings of fallopian tube anomalies in this group of patients has not been well described, though some investigators have suggested that normal, partial, or absent fallopian tubes may exist (8, 9). A wide variety of associated anomalies, including vertebral, renal, and cardiac defects, have been described (11). Ovaries are typically present and function normally. In our own center, in patients with MRKH syndrome undergoing surgical resection of an obstructed rudimentary uterine horn, we have observed that the ipsilateral ovary is

FIGURE 3

A laparoscopic view of a patient with Mayer-Rokitansky-Küster-Hauser (MRKH) syndrome. The ovary is associated with only the distal fimbria of the fallopian tube; the infundibular, ampullary, isthmic, and interstitial segments are absent.



Garrett. Development of fallopian tube fimbria. *Fertil Steril* 2008.

often associated with a fimbrial remnant and no associated ampullary section of the tube (Fig. 3). This has also been seen in women with MRKH syndrome with no uterine structures and just the fimbria associated with the ipsilateral ovary.

Recently, there has been a lot of attention involving the fallopian tube as pertains to carcinogenesis. The fimbriated portion of the fallopian tube has been identified as a preferential site for the formation of early serous carcinomas, supporting the fimbria as a likely site of origin for ovarian epithelial neoplasm (12, 13). Some of the differences in the oncologic potential of the distal tubal segment may be explained by its separate embryologic origin. As we look further into identifying the ultimate relationship of the fimbria to carcinogenesis, our examples of congenital malformations may help to further explain the complexity of fimbrial development and biology.

In addition to the embryologic considerations, the two cases reported herein illustrate that the fimbria can exist independently from the remainder of the tube without dependence on the proximal portion of the tube for blood supply. In both cases the fimbriae were supplied by collateral blood vessels from the pelvic sidewall, and there were no signs of ischemia or atrophy. This implies that procedures such as tubal ligation may not impact the viability of the fimbria, which is an important consideration in light of our new awareness regarding ovarian epithelial carcinogenesis.

CONCLUSIONS

The two cases of congenital fallopian tube atresia that are presented support a novel hypothesis that the fimbria of the fallopian tube may arise separately from the rest of the tube. The observation of the presence of fimbria in women with cervical, vaginal, and uterine agenesis supports this concept. Whether the separate embryologic development can explain the increased oncologic potential of the fimbria remains to be investigated.

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