Review

Müllerianosis

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Summary. Müllerianosis may be defined as an organoid structure of embryonic origin; a choristoma composed of müllerian rests - normal endometrium, normal endosalpinx, and normal endocervix - singly or in combination, incorporated within other normal organs during organogenesis. A choristoma is a mass of histologically normal tissue that is "not normally found in the organ or structure in which it is located" (Choristoma, 2006). Müllerian choristomas are a subset of non-müllerian choristomas found throughout the body.

Histologically, endometrial-müllerianosis and endometriosis are both composed of endometrial glands and stroma, but there the similarity ends. Their pathogenesis is different. Sampson faced the same difficulty with pathogenesis and nomenclature when he wrote: "The nomenclature of misplaced endometrial or müllerian lesions is a difficult one to decide upon." "The term müllerian would be inclusive and correct, but unfortunately it suggests an embryonic origin." Sampson then divided "misplaced endometrial or müllerian tissue" into "four or possibly five groups, according to the manner in which this tissue reached its ectopic location" (Sampson, 1925).

Sampson's classification of heterotopic or misplaced endometrial tissue is based on pathogenesis: 1) "direct or primary endometriosis" [adenomyosis]; "a similar condition occurs in the wall of the tube from its invasion by the tubal mucosa" [endosalpingiosis]; 2) "peritoneal or implantation endometriosis;" 3) "transplantation endometriosis;" 4) "metastatic endometriosis;" and 5) "developmentally misplaced endometrial tissue. (I admit the possibility of such a condition, but have never been able to appreciate it.)" (Sampson, 1925). It is precisely this condition "developmentally misplaced endometrial tissue," [müllerianosis] that is the subject of this review.

Key words: Endometriosis, Müllerianosis, Müllerian choristoma, Pathogenesis, Pathology

Introduction

"The surgeon has a wonderful opportunity to study 'living pathology' in both the early and advanced stages of disease" (Sampson, 1924). When it is realized that Sampson was the sole author of virtually all his publications, we can conclude that he undoubtedly would have recognized and described müllerianosis had he had the cold light laparoscope to study the pelvis. Nonetheless, Sampson did publish an illustration of a "shallow" (peritoneal) pocket in the broad ligament (Sampson, 1927).

Based on his extensive operative experience with cervical cancer, Sampson fully appreciated the invasiveness of endometriosis. In the last sentence of his paper on heterotopic or misplaced endometrial tissue, he concluded: "It would seem that we are warranted in stating that the invasion and dissemination of benign endometrial tissue employ the same channels as the invasion and dissemination of cancer" (Sampson, 1925). Toward the end of his career, Sampson indicated why he introduced the term endometriosis and alluded to the inflammatory reaction associated with endometriotic adhesions.

"The term endometriosis was introduced to indicate the presence of ectopic tissue which possess the histologic structure and function of the uterine mucosa. It also includes the abnormal conditions which may result not only from the invasion of organs and other structures by this tissue, but also from its reaction to menstruation" (Sampson, 1940).

In a monograph dedicated to "Dr. John A. Sampson," the Canadian gynecologist James Robert Goodall described the fourth misplaced or heterotopic tissue – endocervicosis - to complete the benign invasive quartet: adenomyosis, endometriosis, endosalpingiosis,

and endocervicosis. "Endocervicosis is a new disease, a recent discovery. It is characterized by a nonmalignant invasion of the deep cervical and paracervical tissues by the mucosa of the cervix uteri" (Goodall, 1943). Goodall also described specific host responses to the endometriotic stimulus that he observed at surgery and in the pathology laboratory. The host responses to endometriosis included: hypertrophy of the invaded organs, relaxation of supporting ligaments of the uterus and ovaries that permitted uterine retroversion, sclerosis of the ovary and peritoneum of the anterior and posterior pelvic pouches, intense inflammatory response during acute phases of the disease, and the ubiquitous and often extensive adhesions found in chronic phases of the disease. Goodall, Sampson, Cullen, and others before them more often observed advanced stages of endometriosis at laparotomy and at autopsy.

If we consider worse case scenarios encountered with endometriosis and müllerianosis, the reader will immediately appreciate the vast differences between the two conditions. Examples of worse case scenarios for müllerianosis include: two reports of intraspinal choristomas, one an endometrial choristoma (Agrawal et al., 2006), the other a müllerianosis choristoma (Barresi et al., 2006). Both were successfully treated by surgical intervention without complications. A massive endometrial choristoma of the liver was also treated by surgical excision without complication (Tuech et al., 2003).

Every gynecologic and pelvic surgeon has encountered the worse case endometriosis-associated inflammatory scenario – the completely frozen pelvis - with all organs cemented together by dense endometriotic adhesions. For the worse case endometriosis-associated invasive scenario, we refer to the reports of Dr. Thomas S. Cullen. Cullen described a patient who developed postoperative rectovaginal and vesicovaginal fistulas following surgical excision of adenomyomatous growths involving the rectum, vagina and cervix; in effect surgery resulted in a cloaca, a major complication. The surgical specimen is illustrated in Fig. 13, Plate LXXVI (Cullen, 1917). Perhaps there is no more powerful demonstration of the basic phenotypic differences between deeply invasive endometriosis and non-invasive müllerianosis than the devastatingly invasive adenomyomatous growths reported by Cullen and the placid, non-invasive müllerianosis of peritoneal pockets in the floor of the RVPD (Batt et al., 1989).

In his final contribution Cullen concluded: "The removal of an extensive adenomyoma of the rectovaginal septum is infinitely more difficult than a hysterectomy for carcinoma of the cervix" (Cullen, 1920). Later, referring to a case of adenomyomata of the rectovaginal septum that he had seen in consultation after hysterectomy, Cullen remarked: "In this case we found an extension of the growth - an extension so widespread that removal of the adenomyomatous growth was out of the question" (Cullen, 1925). In sum, invasion is the sine qua non of all endometriotic disease

(Koninckx and Martin, 1994; Koninckx et al., 1999). Cullen, Sampson, and Goodall described the pathology and explained the pathogenesis of four phenotypes of benign invasive disease: Cullen, adenomyosis; Sampson, endometriosis and endosalpingiosis; Goodall, endocervicosis. In this review we will describe the pathology and the criteria for diagnosis of müllerianosis in all of its histologic and phenotypic variety, and explain how we arrived at the developmental pathogenesis of müllerianosis.

Historical evolution of the theory of pathogenesis of müllerianosis

Our interest in the pathogenesis of müllerianosis was stimulated by two presentations on endometriosis given at the Buffalo Gynecologic and Obstetric Society in 1984, one by Dr. Donald Goldstein from the Adolescent Gynecology Clinic at Boston Children's Hospital and the second by Dr. Donald Chatman from the University of Chicago. Goldstein referred to a pelvic peritoneal pocket as a "Murphy window" while Chatman called it a "peritoneal defect." Since the term "peritoneal defect" implied a deficiency when there was none, we proposed instead the descriptive term peritoneal pocket, which described an organoid structure, and also because the floor of this organoid structure could be grasped and turned inside out for excision.

Practicing at a highly specialized infertility and endometriosis regional private practice in Buffalo, New York, provided us with many cases of pelvic peritoneal pockets, the common form of müllerianosis. Many peritoneal pockets had tiny endometriotic brim nodules. We found only one that contained an endosalpingiosis cyst, tethered by a stalk. Though we never saw a case of endocervicosis, we included endocervicosis in the definition of müllerianosis (Batt et al., 1990), having been strongly influenced by Goodall (1943) and our earlier work (Batt and Naples, 1982). This decision was also based on an insight that müllerianosis was developmental and that in time we would observe cases of endocervicosis and more cases of endosalpingiosis.

The insight that pelvic peritoneal pockets might originate during embryonic development came on April 12, 1985 when we first saw a patient with the 'bilateral and symmetrical' pattern of peritoneal pockets in the RVPD. The bilateral and symmetrical pattern suggested rudimentary duplication of the primary müllerian ducts and hence, a developmental pathogenesis (Batt and Smith, 1989). This insight was corroborated by the observation of anomalies in 18/54 (33%) of our patients with peritoneal pockets (Batt et al., 1989). Some patients had more than one anomaly in addition to the peritoneal pocket(s). Specifically, 13/54 (24%) of our patients had medial position of the ureter(s), some associated with a large recess in the broad ligament of sufficient capacity to envelop the ovary and fallopian tube. Anomalies of the primary müllerian system were found in 8/54 (15%) of our patients with pelvic peritoneal pockets, and in

addition half of them had medial positioning of the ureter(s). In seven patients the primary müllerian anomaly involved the fallopian tubes; the eighth patient had müllerian agenesis (Mayer-Rokitansky-Küster-Hauser Syndrome) and medullary spongiosis of the upper pole of the right kidney. Also, finding one patient with a central peritoneal pocket surrounded by an extensive plexus of varicose veins and another patient with a splayed-open uterus-like organoid müllerian structure that occupied the RVPD provided us with further clinical evidence for a developmental pathogenesis. In a prospective study we observed pelvic peritoneal pockets in 27% of adolescent and adult women undergoing laparoscopy or laparotomy for endometriosis (Batt et al., 1997). This growing body of evidence supported our hypothesis that müllerianosis was a developmental entity.

In 1990, we defined müllerianosis "as the presence of remnants of müllerian tissue (endometriosis, endosalpingiosis, endocervicosis) associated with peritoneal pockets localized to the rectovaginal pouch, rectovaginal space, posterior broad ligaments, and pararectal space" (Batt et al., 1990). In retrospect, this was not only an unnecessarily restrictive definition necessitating the presence of pelvic peritoneal pockets but also incorrect nomenclature. The terms endocervicosis, endometriosis, and endosalpingiosis imply invasive disease and in our opinion are inappropriate to müllerianosis.

Pathology and pathogenesis of müllerianosis: Revised developmental theory

With publication of the remarkable case report of a huge hepatic endometrioma (Tuech et al., 2003), we recognized a unique resource provided by such 'virtual referrals' (Batt et al., 2003). By 'virtual referrals' we mean case reports of rare müllerian choristomas. Such 'virtual referrals' provide an opportunity to analyze their pathogenesis and pathology. Also, 'virtual referrals' provide two crucial advantages: not only have the cases been completely evaluated, they also have satisfied peer reviewers before publication.

Since 2003 we have analyzed a critical mass of 'virtual referral' cases which has generated a greater appreciation for the phenotypic diversity of müllerian choristomas. The 'virtual referrals' included such rare cases as endometrial cysts of the liver (Batt et al., 2003, 2006a), endometrial lesions of the sciatic and obturator nerves (Yeh et al., 2004a,b), precoccygeal endometrial cysts (Batt, et al., 2006b), and a case of spinal intradural müllerianosis (Barresi et al., 2006). As we encountered more of these rare polymorphic phenotypes, we broadened our inclusion criteria for the developmental theory to address the pathogenesis of müllerian choristomas in diverse locations within the abdominal and pelvic cavities. And we redefined müllerianosis as a choristoma or an organoid lesion comprising müllerian anlage that has been misplaced during embryologic development. Such müllerian choristomas might contain one, two, or all three mullerian components - endocervix, endometrium, endosalpinx; forms frusta of the cervix, uterus, and fallopian tubes, respectively. In sum, we believe the developmental theory provides a powerful explanation for the pathogenesis of müllerian choristomas wherever they are found.

Müllerian choristomas have been identified in non-müllerian tissues, and non-müllerian choristomas have been identified in non-müllerian tissues and possibly in müllerian tissue. As a more complete inventory of müllerian choristomas becomes available for study, a pattern may emerge giving greater insight into their biologic significance and the developmental dynamics responsible for their misplacement.

Müllerian choristomas in non-müllerian tissues

Endosalpingeal-choristomas have been identified in the urinary bladder (Arai et al., 1999) and the vermiform appendix (Cajigas and Axiotis, 1990). An endocervical-choristoma has been identified in the small intestine (Chen, 2002). Uterus-like choristomas have been found in the small intestine (Peterson et al., 1990) and in the conus medullaris with associated tethered cord (Rougier et al., 1993). A müllerian choristoma has been associated with a case of tethered cord syndrome (Molleston et al., 1991). An endometrial-choristoma has been identified in the lung (Schimizu et al., 1998) and in the pancreas (Lee et al., 2002). Lastly, a patient with symptomatic spinal intradural müllerianosis at the "L2 – L3" level has been reported (Barresi et al., 2006). Histologic examination revealed a 1.9 cm encapsulated smooth muscle nodule containing an "admixture of endocervicosis, endosalpingiosis, and endometriosis." This non-invasive organoid lesion "apparently [originated] from terminal phylum…[having] the gross morphology of a terminal phylum ependymoma." Periodic bleeding from the endometrial component appears to have first given rise to neurologic symptoms and signs of three years duration that ultimately led to the diagnosis of müllerianosis at age 42 years.

Non-müllerian choristomas in non-müllerian tissues

A number of non-müllerian choristomas have been observed in various locations. For example, ovarian choristomas have been identified in the kidney (Levy et al., 1997; Hartigan et al., 2006), renal choristomas in the adrenal gland (Barr and Lorig, 1990), lumbosacral area (Alston et al., 1989; Horenstein et al., 2004), and in the heart (Milliser et al., 1972; Lutzen and Lehmann, 1975). Other types include: a liver choristoma in the heart (Brustmann, 2002), a pancreas choristoma in the lung (de Krijger et al., 2004), a spleen choristoma in the pancreas (Ota and Ono, 2004), central nervous system choristomas in the spinal cord (Chung et al., 1998), neck (Tubbs et al., 2003), and a symptomatic neurenteric choristoma with gastric mucosa in the spine (Kantrowitz

et al., 1986).

Possible non-müllerian choristoma in müllerian tissues. To date, only one case of ectopic thyroid tissue has been reported to have been found in the uterus. The authors advance two explanations for this finding: "metastasis of the thyroid follicular epithelial cells via blood" or "ectopia of the congenital thyroid tissue" (Yilmaz et al., 2005).

Discussion

Considerable difficulty may be experienced in trying to understand the concept of müllerianosis and to distinguish it from endometriosis because both require the presence of glands and stroma for definitive histopathologic diagnosis. This is problematic until one remembers they differ profoundly in phenotype, pathophysiology, and pathogenesis. Both conditions must be viewed in clinical context. Differentiation becomes clearer when one realizes that endometriosis is endometrium shed outside the uterine cavity that invades the outer surface of organs, while müllerianosis is endometrium (and at times also endosalpinx and endocervix) misplaced *within* other organs during organogenesis and is associated frequently with congenital anomalies, often existing in the absence of pelvic endometriosis.

Routinely integrating clinical observations at surgery with histologic examination in the laboratory, we accepted one histologic component - endometrium - as diagnostic of müllerianosis (Batt et al., 1989). Young and Clement (1996) redefined müllerianosis in stricter histologic terms to denote lesions "seen at any site" containing "admixtures of endosalpingiosis, endometriosis, and endocervicosis". This definition required two tissue types, and preferably all three, for the unequivocal pathologic diagnosis of müllerianosis, and to differentiate composite from simple lesions. In effect, Young and Clement questioned our definition of müllerianosis as applied to pelvic peritoneal pockets.

We were unprepared to enter into debate with two renowned gynecologic pathologists regarding the definition and diagnostic criteria for müllerianosis because we had no experience with the rarer forms of müllerianosis they had encountered. Moreover, we believed that their reports of endocervicosis of the urinary bladder (Clement and Young, 1992) and three-tissue müllerianosis of the urinary bladder (Young and Clement, 1996) were consistent with our developmental theory of müllerianosis. In retrospect, our decision to define broadly the pathologic criteria for müllerianosis to include all three müllerian tissue types might seem prescient, though it was not (Batt et al., 1990).

When consulting pathologists are confronted in their laboratory with a histologic specimen and a clinical note, the diagnostic requirement of Young and Clement for two and preferably all three-tissue types (endosalpinx, endocervix, and endometrium) makes perfect sense. Unlike Clement and Young who examined pathology

specimens sent to them from all over North America, our observations were confined to one regional practice located in Western New York, an area shown to have a high ecologic correlation between environmental contaminants and prevalence of endometriosis (Carpenter et al., 2001). The more common form of müllerianosis – peritoneal pockets – was largely seen. We studied them in complete clinical-pathologic context, with frequent pathology consultations in the operating room and clinical consultations in the pathology laboratory. This intense collaboration produced our initial insight.

Müllerianosis presents as rare choristomas within most organs in the abdominal and pelvic cavities with the notable exception of the spleen (Batt et al., 2003). In our opinion, müllerian choristomas - whether they contain one, two, or all three-tissue components - can be diagnosed with certainty when three conditions are met: 1) no evidence of pelvic endometriosis; 2) no direct communications with the endocervix, endometrium, or endosalpinx; and 3) when there is no history of surgery on the reproductive organs. When suspected müllerian choristomas contain two or three müllerian tissue components, we agree with Young and Clement that they constitute definitive diagnostic criteria for the diagnosis of müllerianosis. However, when an endometrial choristoma co-exists with pelvic endometriosis, especially deeply infiltrating endometriosis (Cornillie et al., 1990; Leyendecker et al., 2002; Chapron et al., 2006); given our current state of knowledge, diagnosing a müllerian choristoma can problematic.

We agree with Barresi and colleagues that the presence of all three-tissue types (endometrium, endosalpinx, and endocervix) meets the strictest pathologic criteria for the diagnosis of müllerianosis (Young and Clement, 1996), especially when supported by immunohistochemical evidence. We support the authors' speculation regarding pathogenesis, that "embryonic development might give an explanation for müllerianosis occurring in such an unusual site" (Barresi et al., 2006). To be more specific, given the presence of all three histologic components, we postulate that only müllerian tissue from the genital ridge misplaced to the spinal cord during organogenesis fully explains both the pathogenesis and the pathology of this intradural organoid müllerian choristoma.

Conclusions

We encourage vigorous discussion and debate about the definition, phenotypes, pathology, pathophysiology, and pathogenesis of müllerianosis. In future research initiatives aimed at the elucidation of the etiology of endometriosis, we encourage the inclusion of müllerianosis in the research design. We believe that as evidence continues to evolve supporting a multi-factorial etiology for endometriosis, such multi-factorial influences on the embryonic development of müllerianosis should be investigated. While the exact

exposure or its timing remains unknown, research focusing on the effect of xenobiotic agents occurring periconceptually or during early embryonic development may advance our understanding of the biology and clinical significance of müllerianosis. For example, evidence supports an association between exposure to select persistent organochlorine chemicals and risk of endometriosis (Buck Louis et al., 2004; Porpora et al., 2006), but researchers continue to be challenged in determining the timing of exposures that may confer such risk.

At present, "X ray, CT and NMR images cannot differentiate spinal müllerianosis" (Barresi et al., 2006). As imaging technologies continue to be perfected, they may offer researchers precise diagnoses in adolescent and adult women without reliance on surgical intervention. This would allow choice of comparison groups for carefully designed multi-center studies. Choice of comparison groups continues to challenge investigators and, undoubtedly, impacts interpretations of the research (Bloom et al., 2006).

Müllerian choristomas containing endometrium generally bleed causing debilitating health problems (Tuech et al., 2003; Barresi et al., 2006). Müllerianosis must be distinguished from malignancy (Young and Clement, 1996; Arai et al., 1999). Müllerianosis presenting as pelvic peritoneal pockets has been associated with pelvic pain and infertility (Batt et al., 1989). In sum, müllerianosis-associated pelvic pain, müllerianosis-associated infertility, and müllerianosis-associated health problems in adolescent and adult women provide sufficient justification for further research of this disorder. Inclusion of müllerianosis in the intensive investigations into the pathogenesis and pathophysiology of endometriosis and adenomyosis can, in our opinion, provide a more comprehensive understanding of endometriotic diseases and contribute to understanding of developmental disease processes. Thus we believe the diagnosis, pathology, pathogenesis, treatment, and long term management of patients with müllerianosis are worthy subjects for discussion and debate at the Tenth World Congress on Endometriosis in Australia in 2008.

Multidisciplinary approaches to müllerianosis are needed, in particular review and evaluation of information from a registry of 'virtual referrals'. Müllerian choristomas need to be identified in clinical populations, including their locations throughout the abdominal and pelvic cavities of individuals identified. Finally, testable hypothesis should underlie research involving women and primates to assess the fundamental molecular processes in the development of müllerianosis. From an etiologic perspective, comparative sociodemographic profiles and tissues from women and adolescents with müllerianosis should be studied taking into account environmental as well as genetic influences. From a clinical perspective, physicians need training to recognize müllerian as well as non-müllerian choristomas encountered during imaging scans, surgical explorations, and pathologic examinations.

In conclusion, we define müllerianosis as an organoid structure of embryonic origin; a choristoma composed of müllerian rests - normal endometrium, normal endosalpinx, and normal endocervix - singly or in combination, incorporated within other normal organs during organogenesis. Composite müllerian choristomas represent forms frusta of the cervix, uterus, and fallopian tubes, respectively. We postulate further that all müllerian choristomas, including pelvic peritoneal pockets, have a developmental origin. In our opinion, the pathogenesis of müllerianosis is fundamentally different from the benign invasive quartet: adenomyosis, endometriosis, endosalpingiosis, and endocervicosis.

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