Case Report

Hymenal Stenosis and Fibrosis in Two Adult Women

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BACKGROUND: Hymenal abnormalities are most commonly a result of incomplete apoptosis of the urogenital sinus during embryology. Infrequently, however, noncongenital abnormalities of the hymen can occur that can cause significant sequelae such as severe introital dyspareunia.

CASES: We report on two adult women who developed severe introital dyspareunia secondary to hymenal stenosis and fibrosis in the absence of other vulvovaginal pathology. Neither woman had point tenderness of the vulvar vestibule, but their symptoms of searing pain on vaginal penetration was reproduced by stretching the hymen with two fingers. In both cases, conservative treatments with vaginal dilators in combination with topical hormonal therapy failed to relieve their symptoms, but both women were subsequently successfully treated with hymenectomy.

CONCLUSION: Hymenal stenosis and fibrosis can develop in the absence of identifiable vulvar dermatoses. If conservative treatment with topical hormonal therapy and vaginal dilators is unsuccessful, hymenectomy can restore normal coital function.

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H ymenal abnormalities are uncommon in the adult patient. By 13 weeks of gestation, the hymen separates the urogenital sinus from the developing vaginal

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Teaching Point

1. Hymenal stenosis should be included in the differential diagnosis in a woman with introital dyspareunia who does not have tenderness of the vestibule.

lumen. It is formed from the posterior wall of the urogenital sinus and is comprised of both epithelial and vaginal cells. The hymen usually disintegrates in the perinatal period and persists as a thin mucous membrane surrounding the vaginal introitus.¹ Incomplete degeneration leads to congenital hymenal anomalies that usually are diagnosed in adolescence. Infrequently, however, noncongenital abnormalities of the hymen can occur that can cause significant sequelae such as severe introital dyspareunia.

However, noncongenital hymenal anomalies have not been reported in the medical literature (see the "Discussion" section for the details of our literature search). The following cases describe two adult women who developed hymenal stenosis and fibrosis in the absence of other vulvovaginal pathology.

We describe two women with adult-onset hymenal stenosis, which may be an underrecognized cause of secondary dyspareunia in nonadolescent women.

CASE 1

The patient is a 56-year-old woman who presented with a 10-year history of introital dyspareunia after many years of pain-free intercourse. She described the pain as "burning, cutting, and rawness" on penetration. Additionally, she had developed vulvar burning over the previous few years. Her medical history was significant for three cesarean deliveries, a total abdominal hysterectomy for menorrhagia, and 6 years of oral conjugated equine estrogen use. Physical examination revealed a severely erythematous, tender, and atrophic-appearing vulvar vestibule. Thin yellowish discharge was noted in the vagina and there was a loss of rugae of the vaginal mucosa. The hymen was stenotic and, when stretched with two fingers, reproduced the patient's symptoms. A wet mount showed significant leukocytes, hyphae, and parabasal cells and her vaginal pH was 5.5. Her vulvar candidiasis was treated with oral fluconazole and her vulvovaginal atrophy was treated with topical estradiol 0.01% cream applied to both the vestibule and vagina. All evidence of vulvovaginal atrophy had resolved by her 16-week follow-up. However, despite continued use of topical estrogen therapy and the use of vaginal dilators, her hymenal stenosis and introital dyspareunia did not resolve. She

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subsequently underwent a hymenectomy and her dyspareunia resolved. Histopathologic examination of the hymen revealed acute and chronic inflammation and stromal fibrosis (Fig. 1).

CASE 2

The patient is a 49-year-old woman who presented with 8 years of introital dyspareunia after many years of painfree vaginal intercourse. The pain was described as a severe "burning and ripping" sensation on penetration. Prior treatment included vaginal dilators, which provided no relief. She denied nonprovoked pain or burning and found clitoral stimulation to be pleasurable and pain-free. Physical examination revealed a nontender and nonatrophic vulvar vestibule and well-estrogenized, nontender vaginal mucosa. Her hymen was normal in appearance and nontender to touch with a cotton swab; however, the patient's symptoms were reproduced on inserting two fingers into the vagina and spreading them at the hymenal ring. Despite the absence of vulvovaginal atrophy, the patient was treated with a compound that contained estradiol 0.01% and testosterone 0.1% for 3 months without improvement in her dyspareunia. She underwent hymenectomy and her dyspareunia resolved. Histopathologic examination of the hymen revealed subepithelial fibrosis (Fig. 2).

Both patients were diagnosed with adult-onset hymenal stenosis. Both underwent hymenectomy and had complete resolution of their dyspareunia. Hymenal specimens were examined by the George Washington University Department of Pathology, revealing fibrosis.

DISCUSSION

A review of the PubMed and SCOPUS databases was performed using the terms hymen, imperforate



Fig. 1. Hymen. Nonkeratinizing squamous mucosa with acute and chronic inflammation and stromal fibrosis (hematoxylin and eosin, $100 \times$ original magnification). Goldstein. Hymenal Stenosis and Fibrosis in Adult Women. Obstet Gynecol 2015.



Fig. 2. Hymen. Trichrome stain highlights increased subepithelial fibrosis (trichrome stain, $40 \times$ original magnification).

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hymen, hymenal stenosis, hymenal fibrosis, hymenal stricture, and hymenal abnormalities. This database search, which encompassed the years between 1980 and 2014, revealed that the majority of literature pertaining to hymenal abnormalities examines congenital anomalies that are typically diagnosed in adolescence. However, there were two case reports of hymenal stenosis after surgical procedures that involved the hymen and one case report of recurrence of an imperforate hymen during pregnancy.² The paucity of literature discussing postadolescent hymenal pathology might lead to a delay in the diagnosis of hymenal stenosis in older women. The histopathology in both of these patients reveals fibrosis without evidence of any dermatologic pathology. Fibrosis is an unusual finding in hymenal tissue which, like the nonkeratinized squamous endoderm of the vestibule and nonkeratinized squamous mesoderm (mucosa) of the vagina, is normally composed of elastic connective tissue that provides flexibility to accommodate intercourse and childbirth.^{3,4} It is certainly possible that the hormonal changes of (peri)menopause could explain the inflammation and subsequent fibrosis leading to hymenal stenosis in both of these patients. It appears, however, that if the hymen becomes too fibrotic, even topical hormonal therapy and vaginal dilation may not restore normal coital function. Although this is most likely an uncommon cause of introital dyspareunia, hymenal stenosis should be included in the differential diagnosis of a woman who presents with introital dyspareunia

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without point tenderness of the vestibule on physical examination.

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