Bilateral Bartholin’s Gland Hyperplasia Associated with Bartholin’s Gland Cyst: A Brief Case Report

Hyun-Soo Kim • Gou Young Kim
Sung-Jig Lim • Eun-Hee You' Youn Wha Kim2

Departments of Pathology and 'Woman’s Medicine Center, East-West Neo Medical Center, Seoul; “Department of Pathology, Kyung Hee Medical Center, School of Medicine, Kyung Hee University, Seoul, Korea

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Corresponding Author
Gou Young Kim, M.D.
Department of Pathology, East-West Neo Medical Center, School of Medicine, Kyung Hee University, 149 Sangi-dong, Gangdong-gu, Seoul 134-727, Korea
Tel: 02-440-7551
Fax: 02-440-7564
E-mail: pathogen@medimail.co.kr

A 40-year-old woman underwent surgery to remove tender bilateral vulvar masses. The masses were gray/brown, well circumscribed, non-encapsulated, and were composed of an increased number of ducts and acini with a normal lobular architecture and a duct-acinar relationship. This appearance was consistent with Bartholin’s gland hyperplasia (BGH). Bilateral Bartholin’s gland cysts were also associated with BGH. Benign tumors and tumor-like conditions of Bartholin’s gland are uncommon, and only a few cases of BGH have been reported in the literature. Hyperplasia is a rare etiology for an enlarged Bartholin’s gland, and must be distinguished histologically from adenoma.

The major vestibular glands of Bartholin are bilateral, racemose and tubuloalveolar. Each gland is made up of acini composed of simple, columnar, mucus-secreting epithelium and a duct lined by transitional epithelium. Benign tumors and tumor-like conditions of Bartholin’s glands include adenoma, hyperplasia, adenomyoma, hamartoma, leiomyoma, mucinous cystadenoma, cellular angiofibroma, mixed tumors, angiomyxoma, and papilloma.1,2 Adenoma and hyperplasia are two of the less common lesions and a systemic way of distinguishing between them has not been well defined until recently.2,3 Here, we present a case of bilateral Bartholin’s gland hyperplasia (BGH) associated with bilateral Bartholin’s gland cysts (BGCs).

CASE REPORT

A 40-year-old woman presented with a complaint of non-specific discomfort in the perineal region for one month. She had a cesarean delivery 13 years previously and a vaginal delivery without episiotomy. She denied having associated vaginal discharge, dyspareunia, or pruritus. Physical examination revealed bilateral firm and tender masses that measured 3 × 2 cm in both lower aspects of the labia minora. A clinical diagnosis of bilateral BGCs was made, and the masses were excised for analysis. The mass from the left side measured 3 × 2.5 × 1 cm. It was not encapsulated and composed of a yellow/white firm mass of 1.8 × 1.2 × 1 cm and a tan/brown cystic lesion of 1 × 1 × 0.8 cm (Fig. 1). The cut surface showed a pale brownish solid tissue with a whorled appearance attached to a cyst that contained golden yellow gelatinous material. The mass from the right side measured 1.7 × 1.2 × 1 cm and was brown in color without clear encapsulation. Its cut surface had a solid fibrous and whorled appearance. A tan/brown cystic lesion of 1 × 0.7 × 0.7 cm was also identified in the mass from right-side.

These specimens were submitted in their entirety for histological examination by hematoxylin and eosin (H&E) staining.
Selected sections were stained with periodic acid-Schiff (PAS) without diastase digestion, Alcian blue, or mucicarmine. Immunohistochemical studies were also performed on formalin-fixed, paraffin-embedded sections of these tissues using the polymer method. For these studies, we used a monoclonal mouse anti-α-smooth muscle actin (SMA) antibody (1:4,000; clone 1A4; code no. M0851; Dako, Glostrup, Denmark) and polyclonal rabbit anti-human carcinoembryonic antigen (CEA) antibody (1:40,000; code no. A0115; Dako). Microscopic examination revealed bilateral well-circumscribed and non-encapsulated masses containing an increased number of ducts and acini with eosinophilic intervening stroma. A normal lobular architecture and duct-acinar relationship were maintained (Fig. 2). The glands were lined by cuboidal-to-columnar epithelium that contained mucinous material and bland, basally located, regular nuclei. A well-circumscribed cystic lesion abutted each mass. The intraluminal space, containing mucinous material, was lined by flattened transitional epithelium with mucin-containing polygonal histiocytes. These findings were consistent with BGC. The intracytoplasmic and intraluminal mucinous secretions were positive for PAS, mucicarmine, and Alcian blue. The glandular acini and intraluminal mucin-containing spaces of dilated ducts were positive for polyclonal CEA (Fig. 3A). Beneath the acinar cells, there were basally-located myoepithelial cells that were strongly immunoreactive for SMA (Fig. 3B).

DISCUSSION

Benign solid masses of Bartholin’s gland are extremely rare, and specifically only a few cases of BGC have been reported.2,3 The lack of well-defined criteria for distinguishing adenoma
from hyperplasia is, at least in part, due to the rarity of these lesions. The final diagnosis is usually provided by a pathologist, and distinguishing BGH from adenoma requires histological analysis. Adenoma is usually unilateral and presents as a well-circumscribed, hard, mobile, vulvar nodule, with maximum incidence in patients less than 30 years of age. In contrast, BGH presents in older patients as multiple small nodules that are not well circumscribed. The majority of patients with adenoma initially present with complaints of dyspareunia or burning pain in the vagina and are treated for chronic vaginitis. Koenig and Tavassoli reported that 12 of 17 women (71%) had signs or symptoms of BGC. Santos et al. have reviewed 72 cases of Bartholin’s gland lesions including 10 cases of BGH. The BGH lesions measured from 12.5 to 45.0 mm (mean, 23.8 mm) and were solid, tan-colored, slightly firm, and non-encapsulated. Eight patients presented clinically with BGCs, and two presented with vulvar lumps, but BGCs were found adjacent to BGH in only three cases.

Koenig and Tavassoli have proposed a set of criteria for distinguishing hyperplasia from adenoma in which hyperplasia is defined as a proliferation of mucinous acini that maintain the duct-acinar relationship, lack encapsulation, and is either lobulated or has irregular contours. In contrast, the lesion is designated as adenoma if the proliferation of glands, tubules, and acini is haphazard or diffuse, and if the lesion is sharply circumscribed or encapsulated. Our case demonstrated an increase in both glandular and stromal elements, with a normal architectural pattern. Clear demarcation between normal parenchyma and the affected tissue, typically found in adenoma, was not identified. In addition, we recently reviewed 12 cases of BGC and found some differences between the normal-appearing parenchyma adjacent to BGC and hyperplasia of this case. Except for the cystectomy specimen, normal glands adjacent to BGC measured less than 1 cm (0.2 cm to 0.9 cm) and were decreased in number and more sparsely distributed. Closer to the cystic lesion, the glands were atrophic or compressed.

In BGH, periacinar myoepithelial cells stain positively for SMA, and intraluminal and intracytoplasmic mucinous secretions are positive for polyclonal CEA. Our immunohistochemical findings were consistent with those of previous studies.

The treatment of choice for BGH is complete gland excision to exclude malignant neoplasms, such as squamous cell carcinoma, adenocarcinoma, or adenoid cystic carcinoma, which rarely arise in Bartholin’s gland. Follow-up data from reports of benign solid tumors of Bartholin’s gland have demonstrated an excellent prognosis. Our patient likewise showed no evidence of recurrent disease two months after surgery. She has been receiving regular follow-up examinations.

In conclusion, BGH is a rare, benign disease that can be mistaken clinically for several other conditions. BGH should be distinguished from Bartholin’s gland adenoma. Our report highlights clinical and histopathological findings of BGH that can be used for differential diagnosis.

REFERENCES