Case Report

Nodular Hyperplasia of the Bartholin's Gland: An Overlooked Clinical Entity

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Abstract

The Bartholin gland can give rise to several lesions that include: cysts from duct obstruction, carcinomas and much rarely nodular hyperplasia and adenomas. Nodular hyperplasia is an under-reported entity that most often presents as a Bartholin cyst to the gynecologist. We report a case of a 21-year-old Nigerian student who presented with a nontender left cystic vulva swelling with associated vaginal discharges and occasional pains of a month duration. She had an excision of the nodular mass that was seen on the marsupialization of the cyst. The histology of the excised tissue showed nodular hyperplasia of the Bartholin's gland. Tumors of the Bartholin glands are rare in general, with malignant neoplasms easily differentiated from benign ones with histology and clinical presentation. However, benign neoplasms and reactive conditions of the Bartholin glands can easily be confused as both have similar clinical presentations. This distinction is easily made with histology.

Keywords: Bartholin cyst, nodular hyperplasia, vulva lesion

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Introduction

The female genital tract contains several glands of which the Bartholin gland serves to produce lubricating mucus during sexual arousal through the ducts that open at the vulvar vestibulum.[1] Lesions of the Bartholin glands are mostly benign and they are infrequent.[2] The Bartholin gland can give rise to several lesions which may be neoplastic (Bartholin gland carcinomas and Bartholin gland adenomas) and reactive (i.e., Bartholin gland cysts).[3] Bartholin gland lesions that present as predominantly solid lesions of the gland are extremely rare compared to cystic lesions and they are frequently carcinomas.[4] Thus to the clinician, a solid lesion in the Bartholin's gland raises a suspicion of malignancy.[4] Among the benign conditions, the diagnosis of "Bartholin cyst" involving the duct of this gland remains the most common entity clinically diagnosed.[2]

Nodular hyperplasia of the Bartholin's gland is a rare entity which is often mistaken clinically for a Bartholin cyst.[4] Majority of the cases reported in the literature exclude the geographic locale of our practice in Auchi, in the Niger-Delta area of Nigeria. To the knowledge of the authors, this condition – nodular hyperplasia of the Bartholin glands has not been highlighted in recent reviews of female genital lesions within Nigeria.[5-7] Lesions commonly reported from the Bartholin gland in our environment include cysts and abscesses.[5,7,8] Histologically, the first attempt at distinguishing between nodular hyperplasia and adenoma was made by Koenig and Tavassoli.[9] Their description highlighted the irregular or lobulated contours of nodular hyperplasia which is composed of bland mucinous acini with the maintenance of the normal duct-acini relationship.[9]

Presentation is usually as a cystic swelling with or without pain along with dyspareunia.[3,10,11] In addition, the swelling has also been described as increasing in size during sexual intercourse.[10] Some patients can also be asymptomatic and are only discovered during examination for other reasons.[3]

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We present a case of the 21-year-old female student who presented with a vulva mass with an accompanying diagnosis of nodular hyperplasia of the Bartholin's gland along with important diagnostic considerations from similarly presenting reactive lesions

Case Report

A 21-year-old female who presented with a left-sided vulva swelling of a month duration with associated pains and mild vaginal discharge. There was positive history for use of emergency contraception (levonorgestrel). She had a positive history of dyspareunia without accompanying bleeding. Her past medical history was unremarkable.

On examination, she was afebrile and had a blood pressure of 130/75 mmHg. Vaginal examination showed a cystic, tender mass located on the left vulva wall measuring 4 cm by 2 cm. There was no differential warmth. The cervix, pouch of Douglas, and uterus were unremarkable. A clinical diagnosis of Bartholin's cyst was made. Surgical excision of the mass and marsupialization were undertaken following patient's consent. Excised tissue was subjected to histological evaluation. The patient was then placed on broad-spectrum antibiotics postsurgery.

Consent was obtained from the patient to share the photomicrographs and other data which are presented in this report. Institutional ethics committee approval was secured for this report.

Histopathology

Macroscopically

The specimen received was a grayish-white fibrous tissue that measured 2 cm × 1.5 cm × 0.4 cm. It looked like the wall of a collapsed cyst with grayish-white nodular thickening of part of its circumference.

Microscopy

Examined sections on histology showed poorly circumscribed lobules of acini which are lined by mucinous cells with bland basal nuclei. There is the preservation of the duct-acini relationship with foci of squamous metaplasia in a few ducts [Figures 1 and 2].

Follow-up

The surgical wounds healed appropriately and the patient has not reported any further swelling or dyspareunia 3 months posttreatment.

Discussion

As reported in literature, our index case was clinically diagnosed with Bartholin cyst, emphasizing the need for histologic distinction between these Bartholin cyst and nodular hyperplasia as clinical presentations of both are similar.[4] Other benign lesions of the Bartholin's gland which could present similarly include cold abscesses, endometriosis, and tuberculosis.[11]

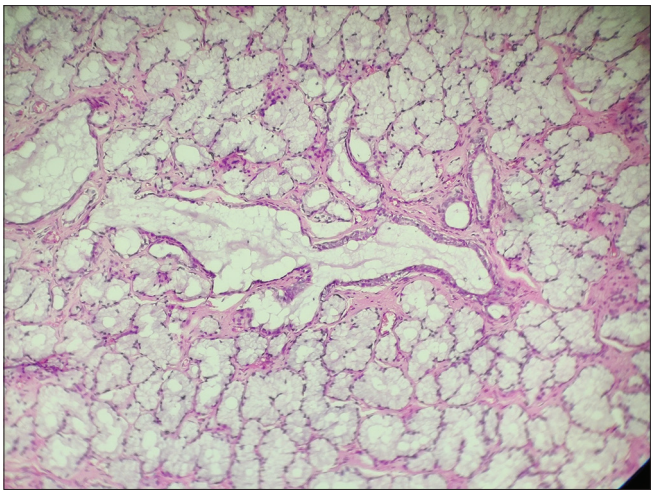

Figure 1: Sections show hyperplastic mucinous glands with preservation of acini-duct relationship (H and E, ×100)

Nodular hyperplasia has a wide age range for presentation, ranging from 19 to 56 years in reported cases.[9,12,13] Our patient was a 21 year old student. The lesions could be solid, cystic, or solid cystic.[3,9]

Nodular hyperplasia of the Bartholin's gland commonly presents with dyspareunia and increase in vulva swelling with associated pain following sexual arousal.[1,10]

It is important to differentiate nodular hyperplasia from an adenoma of the Bartholin's gland as the latter is a neoplastic condition while nodular hyperplasia is a physiological adaptive response. This can easily be achieved by applying the criteria established by Koenig and Tavassoli.[9] It should also be noted that there could be extravasation of mucin and could raise suspicion for an aggressive angiomyxoma.[14] In situation of mucin extravasation, special staining technique can be done to differentiate stroma mucin from a myxoid matrix in an angiomyxoma. Mucin would stained diffusely for mucicarmine, PAS and Alcian blue. CD34 and SMA immunohistochemistry can be done to highlight the neoplastic cells in angiomyxoma.[14]

Although nodular hyperplasia is considered to be nonneoplastic, clonality has been demonstrated in some cases.[13] Similarly, James Yahaya, reported a case of Nodular hyperplasia with dysplastic epithelium.[12] This is suggestive that some cases of nodular hyperplasia could actually be neoplastic as against just a proliferative reactive process. It could be argued that clonality is not equivalent to neoplasia as some authors have reported clonal population in a few normal human tissues while some neoplasms have exhibited polyclonality.

In a 10-year review of vulva lesions diagnosed in Calabar, Southern, Nigeria, Omotoso *et al*.[15] only recorded a single Bartholin cyst representing 0.02% of the 46 cases within the reviewed period and adenomyosis was the most common benign lesion of the vulva in the review.[15] The Calabar study was hospital based, conducted in the pathology department

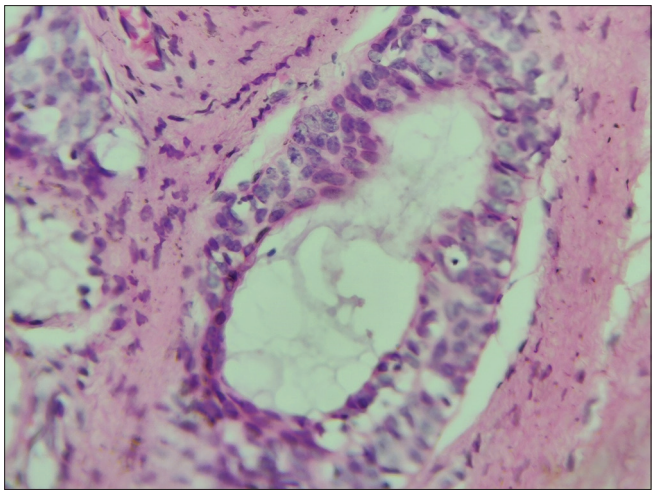

Figure 2: Photomicrograph showing Bartholin glands with focal squamous metaplasia of a duct (H and E, ×400)

with a possibility that many cystic lesions of the Bartholin gland among the wider population were not captured.

The pathogenesis of nodular hyperplasia is unknown but has been related to development of Bartholin ducts cysts which usually follow postinflammatory duct obstruction within the Bartholin gland system.[16] There is local risk of recurrence, when this lesion is incompletely excised.[16] The difficulty in complete excision is due to the relative small size of the Bartholin's gland. The mean diameter of excised glands in a case series was 2.3 cm with a range of 1.2–4 cm.[1] This index case was 2 cm in the widest diameter.

Conclusion

An accurate diagnosis of nodular hyperplasia follows a thorough clinical and histopathologic examination. Benign neoplasms and reactive conditions of the Bartholin glands can easily be confused as both have similar clinical presentation but can easily be differentiated on histology. Bartholin gland hyperplasia remains a rare condition but may possibly be under reported in our environment.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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