



A Bartholin's gland with nodules and cysts bathed in mucus



Matthé P.M. Burger^{a,*}, Concetta M. Salvatore^a, Maaïke C.G. Bleeker^b

^a Departments of Obstetrics and Gynaecology, Academic Medical Center, University of Amsterdam, The Netherlands

^b Departments of Pathology, Academic Medical Center, University of Amsterdam, The Netherlands

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ABSTRACT

The most common pathology of the Bartholin's gland is with its draining duct. Marsupialisation or fistulisation are the most common therapies. In some women, the clinical problem is with the Bartholin's gland itself, being hyperplasia with or without duct rupture in the subcutaneous tissue. Presenting symptoms and signs of nodular hyperplasia are pain and (nodular or diffuse) swelling especially during sexual arousal, or simply a symptomless mass. During surgery, the striking features are subcutaneous free-floating mucus and nodular enlargement 'deep' in the labium majus. The optimal surgical approach is an excision through an incision alongside the labiocrural fold.

We describe three cases to provide insight into the various clinical presentations and management problems. Nodular hyperplasia of Bartholin's gland is easily misdiagnosed because of its rare occurrence and diversity of signs and symptoms. An unusual presentation or clinical course of disease may indicate nodular hyperplasia.

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1. Introduction

The Bartholin's gland is situated deep in the posterior part of the labium majus, measuring 8–10 mm in diameter and has no capsule. Its function is to produce mucus as a lubricant upon sexual arousal. The secreted mucus empties into the Bartholin's duct which measures approximately 2.5 cm in length and opens in the vulvar vestibulum halfway the posterior quadrant, just distal of the carunculae hymenalis.

The most common pathology results from the obstruction and subsequent dilatation of the duct. The usual therapy is marsupialisation or fistulisation (e.g., with the Word catheter), resulting in a new junction between the duct and the vestibulum. Occasionally, the surgery has an unexpected twist when the cyst turns out to be a nodule or no duct wall can be found. In other women, surgical interventions are ultimately ineffective. We present three case histories that each show different aspects of a clinical problem with the Bartholin's gland itself: nodular hyperplasia.

2. Case Histories

Patient no 1 was 25 years of age when she consulted a gynaecologist because of a swollen labium majus on either side. She has pain during and after sexual arousal. The swelling of the labia majora was diffuse and soft, reminiscent of non-pitting oedema, and no nodes were

palpable. The legs and pelvis were thin. The MRI showed a multicystic structure with a diameter of almost 3 cm on either side laterally in the perineal area. In addition, a cystic structure of more than 6 cm length was observed in the right labium majus directly underneath the skin [Figs. 1 and 2]. The gynaecologist decided to conduct a surgical exploration. Directly upon the incision of the vestibulum, an abundant amount of clear mucus flowed off. No cystic wall could be identified. A nodule was palpated both left and right at the lower medial aspect of the inferior ramus of the os pubis, and subsequently excised. The wounds were primarily closed and healed uneventfully. The pathologist diagnosed glandular hyperplasia with mucus outside the glands. Six weeks later, at the post-operative visit, the patient complained of ongoing swelling and pain upon sexual arousal on the right side of the vulva. Upon inspection, the right labium showed a prominent contour with a diffuse and soft consistency underneath, apparently as a result of subcutaneous mucus [Fig. 3]. Upon deep palpation, a nodule of 2 cm diameter was identified. In a repeat procedure, with an incision through the labio-crural fold, the mucus was drained and the nodule excised [Fig. 4]. Histology again showed nodular hyperplasia [Fig. 5]. Postoperatively, the patient went through a period of vestibulodynia at the right side.

Patient no 2 was a 28-year old woman with a recurrent painful swelling of the left labium majus for three months. A course of antibiotics provided only temporary benefit. On clinical examination, the contours of the labia were normal. The gynaecologist found a 'rather deeply' situated soft round swelling of 1.5 cm diameter on the left side. The aim of the surgical intervention was a marsupialisation. However, there was no cyst as expected; instead, there was a rather solid elastic swelling on the lower medial aspect of the inferior ramus of the pubic bone. The swelling was partly excised. The pathologist described the presence of

* Corresponding author at: Department of Obstetrics and Gynecology, Academic Medical Center, University of Amsterdam, Meibergdreef 9, 1105 AZ Amsterdam, The Netherlands.

E-mail address: info@mpmburger.nl (M.P.M. Burger).

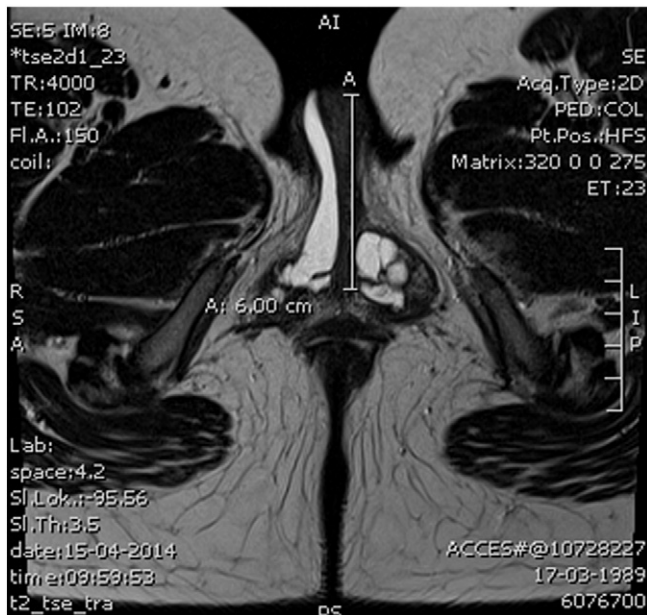


Fig. 1. Pelvic MRI of patient no. 1. Bilaterally cystic structures in or around the Bartholin's gland and a large accumulation of 'liquid' in the stroma of the labium majus at the right side.

gland tissue and myxoid changes in the stroma matching accumulation of mucus. The patient had a protracted postoperative course with bloody discharge, pain and swelling of the left labium majus. Antibiotics did not change her clinical condition. She gradually recovered. Four months after surgery, the patient complained of a painful swelling that was egg-sized on the left labium majus during sexual arousal. Upon clinical examination, the left labium majus was slightly larger than the right one, and its skin is hyperalgesic. Compression of the labium produced clear mucus in the vestibulum. Laterally in the perineum, a swelling of 1–1.5 cm diameter was palpated. Thirty months after surgery, she did not experience vulvar swelling upon sexual arousal anymore, but there was an abundant flow of mucus during and after



Fig. 2. Pelvic MRI of patient no. 1. Sagittal view through right labium majus showing 'liquid' in the stroma.



Fig. 3. Aspect of the vulva of patient no. 1, before the second surgical intervention. Prominent contour of the lower medial quadrant of the right labium, caused by accumulation of subcutaneous mucus.

sexual arousal. The patient did not opt for repeat surgery. We recently asked the pathologist to re-examine the slides resulting in the confirmation of nodular hyperplasia of the Bartholin's gland.

Patient no 3 was a 23-year-old woman who consulted the gynaecologist because of vulvar pain upon sexual arousal for the prior one and a half years. Upon examination, she showed a swelling of 1.5 cm diameter of the Bartholin's gland, which was painful upon compression. A bilateral marsupialisation was planned. After excising a small circular area of vestibular epithelium, the swelling was shown to be solid. A biopsy was taken for diagnostic purposes, and the surgery was completed. The pathologist observed normal mucinous gland structures without the signs of nodular hyperplasia. In a repeat surgery through a vestibular incision, the mobile and deeply situated nodular glands of Bartholin, measuring 2–3 cm, were excised. The blood loss amounted to 150 ml. The pathologist provided the diagnosis of nodular hyperplasia. No remarks were made of mucinous dissection.

3. Discussion

These three women all had nodular hyperplasia of Bartholin's gland. In nodular hyperplasia, there is an increased number of secretory acini but a normal architectural relationship of the acini and the draining duct [1]. The outer contour of the gland is lobulated or irregular. In older reports, the terms 'adenoma' and 'hamartoma' have been used for lesions that now are designated to be 'nodular hyperplasia' [2,3,4,5]. Reports with a histopathological perspective

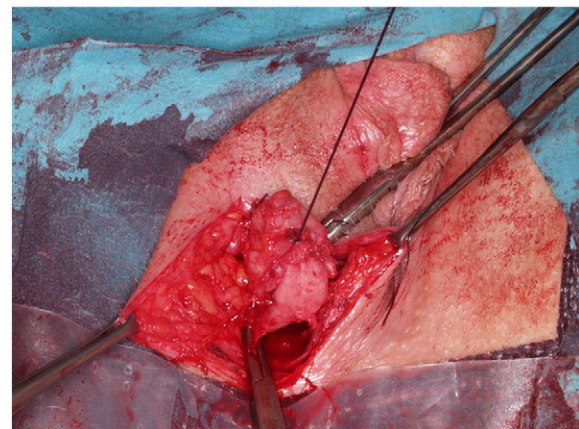


Fig. 4. Patient no. 1. Transcutaneous incision through the labiocrural fold. Nodule of Bartholin's gland is excised.

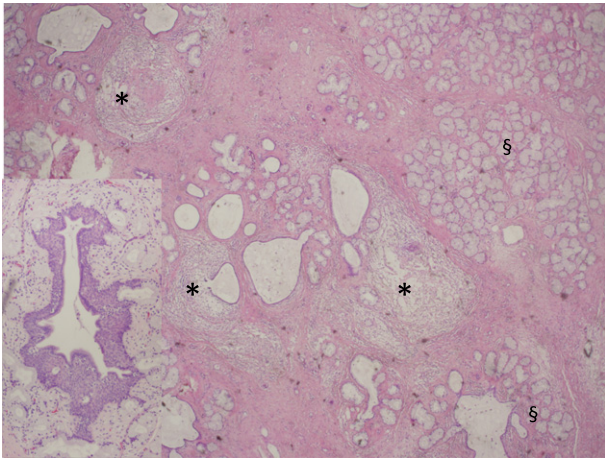


Fig. 5. Nodular hyperplasia at low power of casus 1. There is an increased number of glands with a preserved architecture with numerous acini lined with columnar epithelium (§). The secretion of the acini empties into the Bartholin's duct. In nodular hyperplasia, destruction of the glands with coexisting inflammation, free mucus and mucus-laden macrophages in the stroma (*) and squamous metaplasia (inlet) are often observed.

on nodular hyperplasia are scarce; we counted two larger series with 17 and 10 patients each [1,6] and five case reports with a total of 12 patients [2,7,8,9,10]. The excised glands, as offered to the histopathologist, had a mean maximum diameter of 2.3 cm (range 1.2–4 cm) in the report on 17 patients [1] and 2.4 cm (range 1.3–4.5 cm) in the report on 10 patients [6]. Bilaterality was reported in 1 out of 27 patients in the larger case series [1,6] and in 5 out of 12 patients in the case reports [2,7,8,9,10]. Microscopically, an inspissated mucin and duct rupture with free mucus and mucus-laden macrophages in the stroma has been observed in the majority of the lesions. Disorders of the gland and duct may coincide. Bartholin's duct cysts were reported in 3 out of 10 patients in one report [6] and 2 out of 5 patients in another [1].

The literature on clinical features are even scarcer than on histopathological features. Part of the explanation of its scarcity may be that clinicians have been confused by the clinical presentation and have adopted unusual policies. In the English language literature with a clinical gynaecological perspective, we found 8 case reports of 7 patients; one patient was the subject of two reports [5,11]. The presenting symptoms were pain and swelling with sexual arousal in one patient [4], swelling without pain with sexual arousal in one patient [12], dyspareunia in one patient [13], pain and swelling [14], and no symptoms but a mass in three women [3,11,15]. The mass was bilateral in three patients [4,5,15] and unilateral in four [3,12,13,14]. In the histopathological reports, duct rupture was mentioned in four patients [3,4,5,12], while no attention to this feature was given in three [13,14,15].

It has been suggested that pain and swelling are caused by engorgement of the vascular bed [4]; alternatively, the draining ducts may be stressed as the mucus moves upward from sexual arousal. Pain in cases of occluded Bartholin's gland duct are a well-known phenomenon [16]. In nodular hyperplasia, the volume or consistency of the mucus possibly causes the draining disorder. It is reasonable to assume that women having pain with sexual arousal will also have dyspareunia. In addition, our patients no. 1 and 2 developed *post aut propter*, a vulvar pain disorder, a phenomenon that has been described in the first report on pain during the sex response [16].

In some patients, localized swelling is observed in the area of the lower labium majus. Other patients present with an enlarged labium majus with an exceptionally soft consistency. Such a labium may be filled with mucus, as in our patient no. 1. The mucus is dispersed in the stroma, a condition called 'mucinous dissection'. A nodular enlargement of the Bartholin's gland is to be expected, although nodular hyperplasia can occur without mucinous dissection (in 4 out of a series of 17

patients) [1]. Nodular hyperplasia may occur on both sides but is symptomatic only on one side (as in patient no. 2). Given the difficulties in making a diagnosis (see below), bilaterality is presumably underdiagnosed.

In patients with nodular hyperplasia, there are several clinical pitfalls. As with patient no. 1, an enlarged gland can be missed because the swelling is found deep in the perineum, 2–3 cm lateral and inferior to the fourchette [3]. This area is at or lateral of the labiocrural fold. At palpation, both the intravaginal index finger and the opposite external thumb must make contact with the bony pelvis before being moved medially and posteriorly. Because the clinician is unfamiliar with enlarged glands, the swelling may be misinterpreted as a 'deeply situated' Bartholin's cyst.

It is possible that the gynaecologist aims to perform a marsupialization but is unable to find the wall of the dilated duct after opening the mucus collection. In that case, it is important to look after an enlarged gland deep in the stroma. Some clinicians took a biopsy and postponed the excision to a later date. In patient no. 3, the biopsy showed a normal tissue architecture, while the later excision was nodular hyperplasia. Indeed, the diagnosis can be missed in small biopsies, given the preserved duct-acinar relationship in nodular hyperplasia. Moreover, the density of the acini may vary as exemplified by comparing the densities of the acini in the upper and lower right quadrants of Fig. 5. In performing the excision, part of the excised nodular gland may be left behind. Our patient no. 1 needed a second operation, and other authors had a similar experience. Part of the problem may be explained by the fact that the gland has no capsule, and it may be difficult to find the surgical dissection plane. In one series, the lesion focally involved the surgical margin in 8 out of 10 patients [6]. On the other hand, the enlarged gland may be deeply situated in the stroma laterally of the labiocrural fold. When the gland is approached through a vestibular incision, a complete excision may be difficult. A better incision, delivering more space and a bigger field of view, is a transcutaneous incision alongside the labiocrural fold. This incision is used in the patient depicted in Fig. 4.

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