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Case Reports

Sexual arousal and nodular hyperplasia of the Bartholin gland: a painful combination for a vulval tumor

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Summary

Background: Nodular hyperplasia causes enlargement of the Bartholin gland. The standard approach for a painful enlarged gland is marsupialization. *Case Report:* A 48-year-old woman presented with vulvar pain, which began suddenly 18 months ago. On clinical examination at that time there was a palpable mass of 2 cm. Due to the persisting pain the mass was incised. Pathology report showed that there was nodular hyperplasia. After a while the mass returned accompanied with pain; however the latter also increased during sexual arousal. Multiple treatments were attempted. At the end, it was decided to remove the mass. Pathology examination showed the complete removal of multinodular mucinous gland structures filled with mucus. The hypothesis is that the mucus blocked the gland and caused pain during sexual arousal. *Conclusion:* The treatment for nodular hyperplasia of the Bartholin gland is excision and not incision.

Key words: Vulva; Mass; Nodular hyperplasia; Bartholin gland; Tumor; Sexual arousal.

Introduction

Vulvodynia or chronic vulvar pain in the absence of objective abnormalities has a lifetime prevalence of almost 30% [1]. The pain can be divided in a localized provoked pain at the vestibule (provoked vulvodynia or PVD) or an unprovoked, diffuse vulvar pain affecting the entire vulva (generalized vulvodynia or GVD) [2]. PVD can be further subdivided in a lifelong or primary PVD (PVD1) and an acquired or secondary PVD (PVD2). PVD1 is pain since the first vaginal penetration and PVD2 is pain after a period of pain-free activities [2]. Despite all classifications, the pain often remains a mystery. This is mainly due to the lack of evidence-based medicine studies. In case of no clinical abnormalities, the pain is often considered a psychological problem. Sexual abuse is often a cause of unexplained vulvar complains. The goal of current article is to describe a vulvar tumor associated with PVD2.

Case Report

A 48-year-old woman was referred for vulvar pain. The pain suddenly appeared 18 months ago and on clinical examination at that time, there was a palpable, smooth and mobile structure of 2 cm close to the tuber ischiadicum. Palpation at that time was not painful. Due to its dorsal location, a Bartholin was almost excluded. The initial treatment consisted of oral contraceptive, amitriptyline, tramadol hydrochloride, and orgametril. During this period, the mass did not grow but the pain remained and she had several MRIs in order to search for an explanation for the pain. On the MRI a more prominent corpus spongiosum could be seen on the right side. Six months after the first clinical visit, an excision biopsy was performed and pathology examination showed a nodular hyperplasia of Bartholin's gland. Over the time the pain increased and the patient could no longer sit straight. Furthermore, sexual arousal increased the pain. During clinical examination, a 2-cm nodule could be palpated between the index finger located in the vagina and the thumb externally. The mass was located 3 cm right from the midline, at the height of the posterior fourchette. Palpation increased the pain. The nodule could be mobilized by applying the index finger against the thumb at the labiocrural fold and drawing it medially away from the tuber ischiadicum. A surgical resection of the nodule was performed. The nodule was 1.5 ×2.0 cm in size and the Bartholin gland and all margins were free. Microscopic examination showed that the Bartholin gland consisted of hyperplastic multinodular mucinous gland structures or nodular hyperplasia (Figure 1). The hypothesis is that the mucus blocked the gland and caused pain during sexual arousal.

Discussion

The Bartholin gland or greater vestibular gland is located in the labium majus, lateral, and inferior of the bulbocavernous muscle. The gland contains a duct with a diameter of 5 mm and a length of 1.5–2 cm. They open into the posterior-lateral aspect of the vestibule at 17.00 (left) and 19.00 (right) o'clock. During sexual arousal, the gland se-

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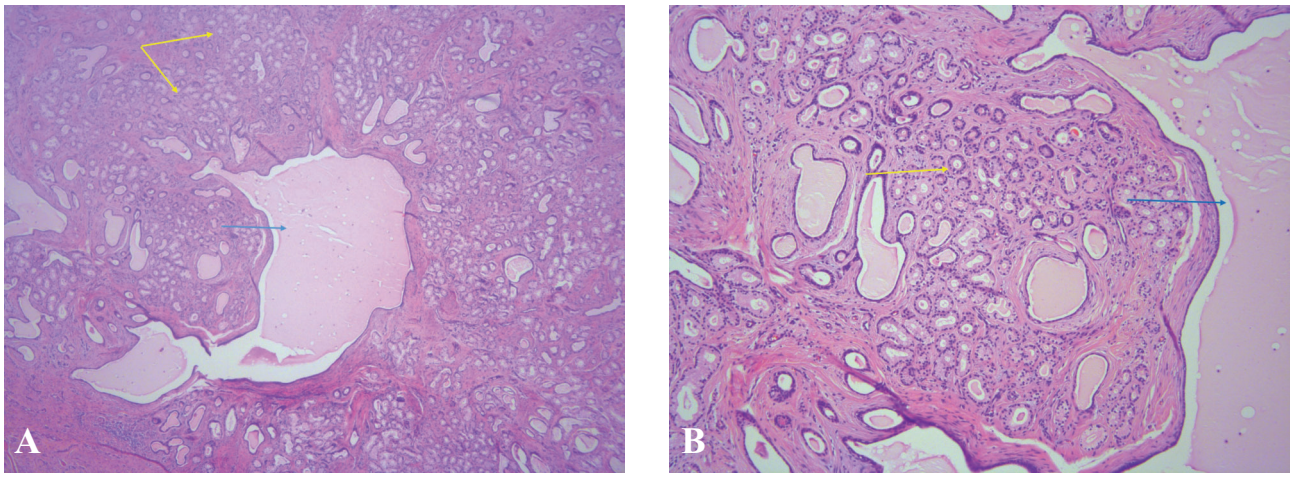

Figure 1. — A and B (enlargement): Central dilated duct (blue arrow), surrounded by hyperplasia of sero-mucinous glands (yellow arrow).

cretes mucus for vulval and vaginal lubrication.

A Bartholin gland can enlarge due to an infection, ductal obstruction, squamous carcinoma, adenocarcinoma, sarcoma, non-Hodgkin lymphoma or a benign tumor [3]. An epithelioid sarcoma, which originates in the deep or superficial tissues of the vulva, is misleading because it can give the impression of an enlarge Bartholin gland [4]. A mesenchymal tumor can also be confusing as it gives a non-ulcerating, painless vulvar mass [5]. Rather surprisingly, nodular hyperplasia is one of the most uncommon forms of Bartholin enlargement with less than 40 cases published in the English literature [6]. A pathognomonic sign for Bartholin duct obstruction by nodular hyperplasia and PVD1 is increasing pain during sexual arousal. Other presenting symptoms reported in the literature are swelling during sexual arousal without pain, pain and swelling, dyspareunia or a painless mass [6-11]. Increasing pain during sexual arousal is caused by the accumulation of the produced mucus in the occluded Bartholin duct. Occlusion of a Bartholin duct after vulvar surgery in the area is relatively high. For instance, after a superficial vestibulectomy, almost one in ten patients will have a duct occlusion and 4% will experience pain and/or swelling during sexual arousal and vestibulectomy [12]. The re-occlusion of the duct or the recurrence rate for a Bartholin cyst or abscess, after marsupialisation or a Word catheter is also 10% [13]. In nodular hyperplasia, the mucus is obstructing the duct. This causes enlargement of the duct and eventually spread of the mucus in the surrounding tissue. The latter is also called mucinous dissection [6]. The location of a Bartholin's cyst and an enlarged Bartholin gland due to nodular hyperplasia is slightly different. A Bartholin's cyst will be more superficial, while the enlarged gland due to nodular hyperplasia will be deeply situated in the perineum, 3 cm lateral and inferior to the fourchette [6, 9].

The treatment of a Bartholin gland with nodular hyperplasia is complete excision. In the past, a complete excision was also the standard approach for a Bartholin cyst or abscess. By this approach, the recurrence rate was zero. The disadvantages included the infection rate, hematoma formation, hemorrhage, decreased lubrication, and mutilation of the vulva. Over time, the surgical approach has changed from a complete removal to the creation of a new duct opening. The morbidity has decreased considerably with a rate of 10% recurrence. The lifetime risk of a Bartholin cyst or abscess is 2%. This high incidence is the reason that every swelling in the Bartholin region is incised and not excised. If a biopsy of the tissue for pathological examination is not taken, then there is risk of misdiagnosis, therefore, it is recommended during the surgical procedure. Sometimes this will offer surprising results like cancer or a benign tumor. In case of a recurrent swelling in the Bartholin region, a thorough re-examination should be performed with a revision of the pathology. Special attention should be given to triggers that cause the complaints. Also, the location of the swelling and the pain intensity are important. If a re-operation is considered, then this could be an incision once again or a complete excision. Nevertheless, in both cases, tissue should be sent to pathology. When nodular hyperplasia is detected, a complete excision should be performed.

Conclusion

A swelling in the Bartholin region is very common, especially in premenopausal women. When the swelling is associated with pain during sexual arousal, a nodular hyperplasia of the Bartholin gland should be considered. Ideally in these cases, one would perform a complete excision. However due to the rareness of this benign tumor, it is rec-

ommended to perform an incision with the creation of a new duct opening. In all cases a tissue sample should be send to pathology. If definitive pathology shows it is a nodular hyperplasia, a complete excision is advised. When despite all precautions, there is recurrence of swelling in the Bartholin region, a thorough re-examination should be performed to exclude an underlining benign or malignant tumor.

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