



Surgical treatment for provoked vulvodynia – Where do we stand? A narrative review

Joana LYRA¹, Joana LIMA-SILVA^{1,2}, Pedro VIEIRA-BAPTISTA^{1,2,3}, Mario PRETI⁴, Jacob BORNSTEIN^{5,6}

¹Lower Genital Tract Unit, Centro Hospitalar de São João, Porto, Portugal

²Department of Gynecology and Obstetrics, Hospital Lusíadas Porto, Porto, Portugal

³LAP, a Unilabs Company, Porto, Portugal

⁴Department of Surgical Sciences University of Torino, Torino, Italy

⁵Azrieli Faculty of Medicine, Bar Ilan University, Safed, Israel

⁶Research Institute, Galilee Medical Center, Nahariya, and Azrieli Faculty of Medicine, Bar-Ilan University, Safed, Israel

ABSTRACT

The treatment of vulvodynia remains challenging. Surgery (vestibulectomy) is an option for localized vulvodynia, but it is often considered only after failure of conservative approaches. The authors reviewed the available literature to establish the role, indications, complications, and success rates of surgical procedures.

We conducted a literature search of all the papers published and indexed in PubMed since 2011 on the surgical treatment of vulvodynia.

Women with localized provoked vulvodynia (LPV) form are the best candidates for the surgical treatment of vulvodynia. Success is associated with secondary LPV, improvement with lidocaine, premenopausal status, and intermittent rather than constant pain. While medical/conservative treatment should generally be the first option, if a neuroproliferative etiology is suspected, surgery can be a first-line treatment. The available data do not allow us to draw conclusions about the best surgical technique. Efficacy (defined in different ways) is high (52%–97%). The complication rate is low, cosmetic results are good, and vaginal delivery seems possible.

Vestibulectomy is a safe and effective treatment for vulvodynia when delivered to appropriately selected women.

Keywords: Neuroproliferation; provoked vulvodynia; surgery; vestibulectomy

INTRODUCTION

In 2015, the International Society for the Study of Vulvovaginal Disease (ISSVD), the International Society for the Study of Women Sexual Health, and the International Pelvic Pain Society published a unified definition of and terminology for vulvodynia. Vulvodynia is defined as vulvar pain of at least three months' duration without a clear identifiable cause that may have potential associated factors.¹

Vulvodynia affects all age groups at a prevalence of 6.1%–20.8%²⁻⁴ in premenopausal women, when this condition appears more frequently.⁵ Besides its recognized prevalence, it is believed to be underdiagnosed, with only 60% of symptomatic women seeking help and 40% of these patients never receiving a diagnosis.⁶ As a chronic disorder, vulvodynia is associated with significant health and psychological burden as well as considerable costs.⁷⁻⁹

Address for Correspondence: Joana Lyra, Lower Genital Tract Unit, Centro Hospitalar de São João, Porto, Portugal

E-mail: joanaritalyra@gmail.com ORCID: orcid.org/0000-0002-3622-3850

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We can further categorize vulvodynia in terms of location (localized to the vestibule or the clitoris, also named vestibulodynia or clitorodinia), generalized, or mixed; provocation (upon contact or spontaneous); onset (primary or secondary); and temporal pattern (intermittent or constant). These descriptors were defined in a more recent document published by the same consensus group.¹⁰ Despite attempts to further categorize vulvodynia, this is a difficult task, as we still lack important information about its etiology. It is considered a multifactorial condition that envelopes a large spectrum of symptoms and pathophysiological events.¹¹ In fact, its diagnosis is one of exclusion. The main advantage of the 2015 consensus terminology is that it leads to a paradigm shift in treatment from a trial-and-error approach to tailoring the treatment according to the associated factors; if a significant musculoskeletal factor is documented during the physical examination, physical therapy is proposed. When a psychological factor is revealed, the treatment should consider it.¹²

The new paradigm has not been studied in large cohorts; therefore, most data published to date on treatment options are of low quality. In addition, the studied treatments were often performed without proper patient selection.^{13,14} In the past, a multi-step approach starting with conservative and medical treatments was advocated.^{5,12} Surgery, since it is a more invasive and definitive option, is usually viewed as a last resort. Surgery should be offered earlier to women with peripheral neurologic associated factors, that is, neuroproliferation. This phenomenon can be a consequence of increased inflammation in vestibule, although the association between the latter and mucosal allodynia remains to be proven.^{11,12} Regarding the inflammatory milieu, the most consistent finding is an increased number of mast cells in the vestibular tissue and a reduced systemic number of natural killer cells,^{9,15,16} which may be triggered by yeast infections.¹⁷ Some genetic polymorphisms can also explain different inflammatory responses to the same insults.^{5,18,19} Inflammation can lead to angiotensin II formation, which induced angiotensin AT2 receptor-mediated neuronal cells, eventually driving nociceptor axon sprouting.²⁰

The present study reviews the literature in the field and summarizes the current state of the art of surgical treatment of vulvodynia.

Materials and methods

We performed a literature search for all studies published and indexed in PubMed using the terms “vulvodynia,” “vestibulodynia,” “clitorodinia,” “surgical treatment,” “surgery,” and “vestibulectomy.” We limited our search to papers and

guidelines written in English for which the full text was available. We included papers published in the last ten years (2011–2021, with 38 articles retrieved). If relevant, older references found in this set of papers were included in the review (the oldest reference included was dated 1983). We included case reports, case series, review articles, randomized control trials (RCTs), systematic reviews, and meta-analyses. All the included studies were performed in humans, and we excluded those performed in animals.

Ethical approval was not required for this study since it was a review of the existing literature.

Considering the new ISSVD classification for vulvodynia, particularly its new approach to diagnosis and treatment, we revisited and reviewed the most relevant literature with a focus on surgery as a treatment option for vulvodynia.

Results

Is there an ideal surgical candidate?

LPV is most likely type of vulvodynia to benefit from surgical treatment. From the recognized factors that are associated with this phenotype, a peripheral neurologic mechanism with neuroproliferation seems to represent the group of patients who are the best surgical candidates.^{12,21,22}

Pure LPV is restricted to a portion of the vulva (vestibule and clitoris) and triggered only by physical contact. It may represent primary or secondary provoked vulvodynia according to the timing of onset at the first contact or after a period of painless contact. The latter classification may be relevant for predicting treatment response since the likelihood of improvement is reportedly higher in cases of secondary vulvodynia.^{10,12,23} This is the most prevalent type of provoked vulvodynia, and its etiology and pathophysiology are the most commonly studied.^{8,13,24}

LPV can be associated with an increased density of nerve endings in the vestibular stroma-neuroproliferation.^{12,25,26} This condition is characterized by an increased density of C-afferent nociceptors in the vestibular mucosa, leading to allodynia.^{12,21,27} The phenomenon of neuroproliferation was recently confirmed by histological examination of vestibulectomy specimens. This study also reported that women with neuroproliferation-associated vestibulodynia treated conservatively presented higher failure rates than surgery.²¹

Furthermore, some factors can help predict the surgical response (Table 1). Secondary LPV has higher success rates than primary LPV.²⁸ LPV that improves with the application of lidocaine is apparently associated with better surgical results;⁹ women who have pain beyond the vestibule (generalized

Table 1. Predictive factors for surgical outcome in treatment of LPV

Predictive factors of surgery results	
Predictors of better surgical outcome	Secondary LPV
	LPV that improves with lidocaine application
Predictors of poor surgical outcome	Generalized vulvodynia
	Postmenopausal woman
	LPV with constant pain
LPV: Localized provoked vulvodynia	

vulvodynia) triggered during a cotton-tipped test or other type of contact and those with severe comorbidities can experience less improvement with surgery;²⁹ surgery in postmenopausal LPV plays a limited role;⁵ and constant pain is associated with a higher risk of surgical failure.³⁰

When should we consider surgery for provoked vulvodynia?

After identifying a possible surgical candidate, the potential timing of surgery must be determined. Considering that surgery is a definitive treatment, in cases in which reservations exist about the pathophysiological mechanisms underlying vulvodynia, most experts would unfortunately recommend proceeding to surgery only once less invasive interventions fail.^{5,22} Surgery boasts similar pain and sexual function outcomes to those of conservative interventions, reinforcing that medical treatment should be considered before surgery.³¹

On the other hand, there is some consistent evidence about the success rates of vestibulectomy for LPV and, since the new 2015 consensus guiding the therapeutic approach considering associated factors, in cases of neuroproliferative LPV without other associated factors, surgery can be considered as an early effective treatment.^{12,32}

Variations in surgical techniques and other practical issues

Anatomical remarks: The vestibule has a ring shape that extends laterally from the hymenial ring to Hart's line localized on the inner surface of the labia minora. It travels anteriorly to the clitoris frenulum and posteriorly to the fourchette. It contains the urethral meatus, vaginal introitus, and vestibular gland openings (Skene and Bartholin's glands). It originates from the urogenital sinus; as such, it has endodermal tissue. It is covered by nonkeratinized squamous epithelium.^{12,18,26} Another important remark is the lower level of estrogen and progesterone sensitivity in the vestibule than in the vagina.³³

Surgical techniques: In 1983, Woodruff and Parmley were the first authors to describe vestibulectomy. It consisted of the excision of a semicircular segment of the perineal skin, posterior

vestibular mucosa, and posterior hymenial ring. Subsequently, the defect was closed by approximation of the undermined vaginal mucosa to the perineum.³⁴

Over the years, new techniques have been described that aim to improve the success rate and decrease complications. The only systematic review that compared the various techniques found no differences in outcomes and complication rates, so the technique should be the one most familiar to the surgeon that allows for the removal the entire painful area.^{5,18}

It is difficult to systematize the different techniques, as few variations exist between them, and most studies did not fully explain all of the surgical steps.

Simplified vestibulectomy: Removing painful areas using a skinning technique and not extending beyond Hart's line. The hymen remains intact.¹⁸

Posterior/modified vestibulectomy: Only the posterior part of the vestibule is excised (from 2 o'clock to 10 o'clock, inside Hart's line). It could be an option for women reporting only posterior pain.^{8,9,12,27} The hymen is not routinely excised. The posterior part of the vestibule is skinned to a depth of 2–4 mm and the hymen is used as a surgical flap with vaginal advancement (1–2 cm) if necessary.

Total vestibulectomy with vaginal advancement: The anterior and posterior vestibules are excised (sometimes used even in the absence of pain in the anterior area to reduce the risk of recurrence), accompanied by a vaginal advancement flap. In addition to removing the parametarial area (1 o'clock to 4 o'clock and 8 o'clock to 11 o'clock), excision extends 5 mm past the hymenial ring all around, resulting in total hymenectomy.³⁵ Posteriorly, the resection area has a wedge shape (from 4 o'clock to 8 o'clock) extending to the perineum 0.5–1 cm beyond Hart's. The vestibule is skinned to a depth of 2–3 mm and removed with the hymen. A vaginal flap is created by dissection of the vaginal mucosa from the rectovaginal fascia with at least 2 cm to allow closure of the defect without tension.

In 2019, Wu et al.²⁶ published a detailed description of surgical technique, advocating extensive vestibule removal, a vaginal dissection sufficient to remove as many pain fibers as possible, and the importance of a tension-free defect closure.²⁶ Still focusing on the technique, other details described in the literature that can facilitate the procedure include the following:^{18,27}

- Use a pen to mark the area that should be removed;
- Catheterize the urethra;
- Inject adrenaline with or without lidocaine solution into the mucosa to improve the hemostasis and postoperative pain;

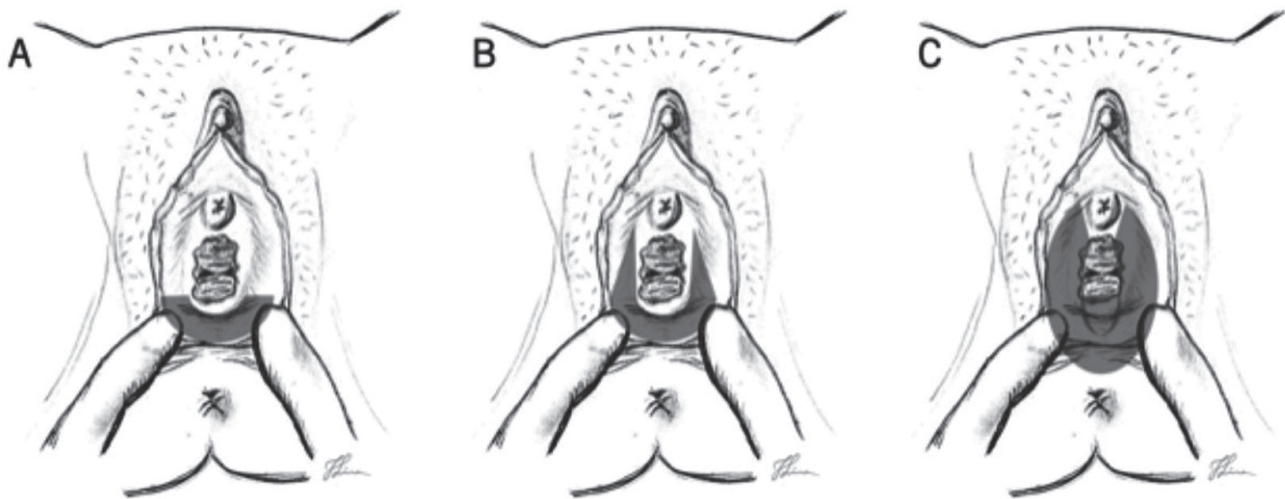


Figure 1. Representation of the three surgical techniques: **A)** Simplified vestibulectomy/vestibuloplasty; **B)** Posterior/Modified vestibulectomy; **C)** Total vestibulectomy with vaginal advancement

- Use two rows of 3-0 Vicryl U-shaped mattress stitches to approximate the vaginal mucosa from the new skin line. The sutures should penetrate the vaginal mucosa backward through the rectovaginal fascia and back again through the vaginal mucosa; they should be placed from anterior to posterior (craniocaudal) to avoid narrowing the introitus. These sutures were not placed anteriorly to avoid urethral injury; and
- Approximate the tissues with interrupted stitches with 4-0 Vicryl and close the anterior defect with a running interlocked 4-0 Vicryl suture.

Postoperatively, the use of ice packs can help prevent swelling and patients should limit physical activity for 4–6 weeks. After complete healing, the use of vaginal dilators could be advocated to stretch the introitus.

Other procedures such as Vestibuloplasty have also been described, but a randomized controlled trial revealed that it failed to improve symptoms.³⁶

Complications: The most frequent complications associated with these procedures are bleeding and consequent hematoma formation, dehiscence, scar tissue formation, increased pain, and Bartholin's gland cyst formation.³⁷ Despite the cosmetic result usually being excellent, there are reports of women unsatisfied with their postoperative vulvar appearance.³⁸ The use of appropriate surgical techniques and some of the precautions mentioned above can diminish the occurrence of these complications. The incidence of immediate postoperative complications seems low.¹⁸ Recurrence rates are variable, ranging from 0% to 13%.¹⁸

The Bartholin's gland openings are adjacent to the hymen at the 5 o'clock and 7 o'clock positions. The occurrence of cysts in this location after vestibulectomy is reportedly 5%–9%, and they

are responsible for pain and bulging symptoms. The need for surgical repair was present in two-thirds of recurrent cases, most occurring from 1.5 months until 2 years postoperative.¹⁸ For this reason, some authors favor the excision of Bartholin's glands during vestibulectomy to prevent postoperative complications.¹² However, this is not consensual, as the risk of cyst formation is low and Bartholin's gland removal could be a complex surgery with increased operative time, blood loss, and risk of pudendal nerve injury.³⁹

Is surgery sufficient or should it be combined with other therapeutic measures?

Despite the ability of surgery to control pain, other concerns persist, and associated symptoms can remain undertreated. For example, there could be decreased vulvar sensitivity after surgery that can negatively affect sexual function. In addition, the prolonged cycle of pain can lead to other psychological dysfunction that limits the full experience of the new painless state achieved with surgery. A combination of physical and psychosexual therapy can amplify the improvement experienced by these patients.^{9,12,32,40} In addition, neuropathic pain medication has been continued postoperatively to maximize the surgical results, both in terms of quality of life and sexual function.⁴¹

Success rates and evidence supporting surgery

Surgery is the most studied treatment for LPV.⁵ Vestibulectomy has a success rate of 65%–90%^{12,42,43} (Table 2), with an 85% likelihood of permanent improvement when performed by an experienced surgeon in selected patients.²⁹ The use of different variations of the same procedure does not seem to impact the global surgery success rate.¹⁸ The outcomes evaluated in previous studies were dyspareunia (improvement in 79–89%),^{18,44}

Table 2. Summary of the studies, surgical techniques, and surgical success rates

Surgery and study	n	Study design	Outcomes	Success rate
Simplified vestibulectomy/vestibuloplasty				
Bornstein, 1995 ³⁶	21	Randomized control trial	Self-reported symptoms	0/10 vs 9/11 (perineoplasty)
Goetsch, 1996 ⁵³	12	Case series (prospective)	Self-reported dyspareunia	83%
Lavy, 2005 ⁵⁴	59	Case series (retrospective)	Self-reported dyspareunia	73.6%
Goetsch, 2007 ⁵⁵	111	Case series (retrospective)	Self-reported dyspareunia	64%
Goetsch, 2008 ⁵⁶	119	Case series (retrospective)	Self-reported dyspareunia	68%
Goetsch, 2009 ³⁹	155	Case series (retrospective)	Vestibular tenderness (touch test)	83%
Posterior/modified vestibulectomy				
Kehoe, 1999 ⁵⁷	57	Case series (prospective)	Self-reported dyspareunia	61.1%
Gaunt, 2003 ⁵⁸	42	Case series (retrospective)	Pain score (objective, subjective findings)	90%
Eva, 2008 ⁵⁹	110	Case series (retrospective)	Self-reported dyspareunia	83%
Goldstein, 2006 ⁶⁰	104	Case series (retrospective)	Patient satisfaction	93%
Tommola, 2011 ⁶¹	70	Retrospective cohort study	Patient satisfaction	91%
Tommola, 2012 ³¹	39	Case-control study	Patient satisfaction	89%
Total vestibulectomy				
Bergeron, 1997 ⁶²	38	Case series (retrospective)	Self-reported dyspareunia	63.2%
Bergeron, 2001 ⁴⁵	78	Randomized control trial	Pain and sexual function	68.2%
Schneider, 2001 ⁶³	54	Case series (prospective)	Self-reported dyspareunia	83%
Rettenmaier, 2003 ⁶⁴	27	Case series (retrospective)	Self-reported dyspareunia	62%
Traas, 2006 ⁶⁵	126	Case series (retrospective)	Pain and sexual function	Sustained improvement
Bergeron, 2008 ⁴⁶	51	Prospective cohort study	Self-reported dyspareunia	52%
Bohm, 2008 ⁶⁶	67	Case series (retrospective)	Self-reported dyspareunia	56%
Bornstein, 2020 ⁵¹	32	Retrospective cohort study	Pain, sexual function, satisfaction	87.5%–97%

n: Number

pain and tenderness of the vestibule evaluated with the cotton-swab test (improvement in 70%–85%), patient satisfaction with the procedure (90%),¹⁸ and improved sexual function.²¹

An RCT of the treatment of provoked vulvodynia that compared surgery with electromyography biofeedback or cognitive-behavioral therapy, reported greater pain reduction with vestibulectomy,⁴⁵ but no differences in dyspareunia were reported after surgery versus cognitive-behavioral therapy at 2.5-year follow-up.⁴⁶

Vestibulectomy failure may be associated with preservation of the anterior part of the vestibule since sensitivity may develop there later with the use of techniques such as posterior/modified and simplified vestibulectomy.¹²

Despite the considerable evidence of surgical success reported by case series and RCTs, well-designed comparative studies incorporating a larger number of patients with LPV selected for surgery are lacking. Studies that use a precise definition of the diagnosis and associated factors, provide a detailed description of the technique used, detail the patient selection criteria, and include adequate follow-up are of major importance for validating future results.^{24,27,47} The need for agreement about validated outcome measures is of the utmost importance before researchers continue to investigate the therapeutic effects for LPV.^{23,48}

Pregnancy and delivery after vestibulectomy

There is little evidence about pregnancy and the best delivery route after vestibulectomy. Burrows et al.⁴⁹ reported that, among 44 women with at least one term pregnancy after vestibulectomy, 21 had a cesarean section and 23 had a vaginal delivery. Among the latter, 48% (11) had no perineal lacerations, 13% (3) required a midline episiotomy, and 4.4% (1) experienced a fourth-degree laceration. The authors considered that vaginal delivery after vulvar vestibulectomy seems safe with perineal morbidity similar to that of the general population. In addition, it alone is not an indication for cesarean section.⁴⁹

According to Bornstein, perineal tears during delivery after vestibulectomy are rare. This may be due to excision of the perineal tissue during the procedure, which removes tension from these tissues.¹²

CONCLUSION

Our knowledge about vulvodynia has evolved, and we have gained a new perspective the past few years, mainly after the revised ISSVD consensus terminology and classification were published in 2015.¹ However, considering the relatively recent systematic approach to the classification of this disease, level 1

evidence is still lacking, the availability of which would provide better insight into the best treatment options, with surgery being no exception.¹⁴

Since we still poorly understand the mechanisms of vulvodynia, it is not simple to rely on its pathophysiology for choosing a treatment option. When we can easily identify a trigger or reason for the pain, treatment will focus on its correction. However, most clinicians will adopt a stepwise methodology, with non-invasive medical treatments coming first and surgery being reserved for refractory cases.^{5,14,31,50} However, not all experts agree that surgery should be considered a last resort. For example, in the specific case of vulvodynia associated with neuroproliferation of the nerve endings in the vestibular endoderm tissues¹ – a recognized alteration that leads to provoked vulvodynia – surgery may be the only indicated treatment.¹² Successful rates range from 52%–97% depending on the series and the most frequent complications are bleeding and consequent hematoma formation, dehiscence, scar tissue formation, increased pain, and Bartholin's gland cyst formation. There is no consensus or sufficient evidence on the best technique, so it should be used the one most familiar to the surgeon that allows for the removal the entire painful area. Overall, surgery is considered a safe and successful option for vulvodynia treatment, mainly when performed in selected women. Furthermore, symptomatic improvement after surgery is maintained for long periods, and in cases where symptoms do not immediately subside after surgery, it is described a gradual improvement over the years.⁵¹

This paradigm shift, in which the treatment choice is determined by the identified associated factors rather than a standardized methodology that fits all women with vulvodynia, is mainly a consequence of the 2015 ISSVD consensus and our best understanding of vulvodynia.^{12,21}

Despite the controversies discussed above, there is consensus about the importance of involving patients in the treatment steps and goals. In most cases, there is no single treatment that will completely resolve symptoms, and it may take time for the patient to experience significant relief.^{12,52} Therefore, discussing and explaining treatment goals and expectations is essential for patient compliance and satisfaction.

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Ethics

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