PERSPECTIVE

Vulvar lichen sclerosus in pregnancy: Unaddressed needs and systematic review

Britt Debaene¹ I Sander Vandeweege¹ Hans Verstraelen^{2,3}

¹Faculty of Medicine and Health Sciences, Ghent University, Ghent, Belgium

²Department of Obstetrics & Gynaecology, Ghent University Hospital, Ghent, Belgium

³Department of Human Structure and Repair, Faculty of Medicine and Health Sciences, Ghent University, Ghent, Belgium

Correspondence Hans Verstraelen. Email: hans.verstraelen@ugent.be

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Dear Editor, vulvar lichen sclerosus (vLS) is a chronic inflammatory skin condition that may affect the anogenital area in women of any age. Treatment of vLS with high-potency topical corticosteroids (TCS) and emollients/moisturizers should be assumed for life, regardless of symptoms, and further adjusted to disease activity.¹ Apart from providing symptomatic relief, long-term maintenance therapy is key in maintaining disease remission, in reducing the risk of scarring and loss of vulvar anatomy, and, notably, in minimising the risk of squamous cell carcinoma in vLS.^{2,3}

In clinical practice we perceive a defined patients' needs however, for information on disease course in relation to pregnancy and childbirth. Obstetric health care providers in turn may not be familiar with vLS. We therefore listed a set of topical patients' questions and concerns, and further sought to provide answers through a systematic literature search in line with the PRISMA statement,⁴ and according to a preregistered protocol (Prospero CRD42023411358).⁵ Specifically, we searched MEDLINE, EMBASE, and Web of Science, without limits or language restriction, up to 12 February 2023 and further cross-checked reference lists of selected studies. Studies were considered eligible if these addressed at least one of the following questions: Does pregnancy alter the disease course of vLS?; Does or may vLS necessitate delivery by primary caesarean section?; Is there an increased risk for perineal tears with vLS?; May vLS delay or complicate healing of episiotomy and/or perineal tears?; and, does vLS worsen following delivery? Eligibility screening of records retrieved was performed independently by all authors. Risk of bias was assessed through use of the appropriate JBI tool for the respective study designs.^{6,7}

In this manner, we retrieved eight studies, published between 1991 and 2021, with data on pregnancy and/or delivery in a total of 177 vLS patients (sample size range, 2–92) (Table 1). Except for a single nested casecontrol study, all studies were uncontrolled and except for one prospective study, all studies had a retrospective case series design.

Both worsening of vLS, not otherwise specified (reported in two studies, with worsening in 5 of 11, and 2 of 8 patients, respectively), as well as improvement during pregnancy (reported in three studies, with improvement in 5 of 11, 3 of 9, and 4 of 4 patients, respectively) was reported, while absence of an observed effect of pregnancy on vLS was by far the most common outcome across studies (reported in four studies, with an absence of effect on vLS in all 33 pregnancies in 29 patients, in all 36 pregnancies in 22 patients, and in 5 of 9, and 6 of 8 patients in two previously listed studies, respectively). Overall, disease course of vLS during pregnancy was reported in 7 out of 8 studies, relating to a total of 85 women, of which 12 patients experienced improvement, 65 women had

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Overview of stu 1991 ¹⁰	idies on vulvar lic Country USA	chen sclerosus in women giving bi Study design Retrospective, case series	irth. Sample size N = 11	Methods Self-report (questionnaire)	Outcomes Disease course
95 ¹¹	NSA	Retrospective, case series	<i>N</i> = 9	Self-report (questionnaire)	Disease course; delivery mode
al. 1999 ¹²	NSA	Retrospective, case series	N = 2	Clinical chart review	Disease course
t al. 2008 ¹³	Germany	Retrospective, case series	<i>N</i> = 4	Clinical chart review	Disease course
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TABLE 1 Overview of stu	dies on vulvar lic	chen sclerosus in women giving bi	irth.		
Study	Country	Study design	Sample size	Methods	Outcomes
Helm et al. 1991 ¹⁰	NSA	Retrospective, case series	<i>N</i> = 11	Self-report (questionnaire)	Disease course
Dalziel 1995 ¹¹	NSA	Retrospective, case series	N = 9	Self-report (questionnaire)	Disease course; delivery mode
Haefner et al. 1999 ¹²	NSA	Retrospective, case series	N = 2	Clinical chart review	Disease course
Günthert et al. 2008 ¹³	Germany	Retrospective, case series	<i>N</i> = 4	Clinical chart review	Disease course
Higgins et al. 2012 ¹⁴	NK	Retrospective, nested case-control study	N = 92 (compared to 66 controls)	Self-report (questionnaire)	Delivery mode; perineal tears and healing
Nguyen et al. 2018 ¹⁵	Australia	Retrospective, case series	N = 29 (33 pregnancies)	Clinical chart review	Disease course; compliance, delivery mode; perineal and abdominal scar healing
Trokoudes et al. 2019 ¹⁶	UK	Prospective, case series	N = 22 (36 pregnancies)	Self-report	Disease course; delivery mode; episiotomy and perineal tear rates
Kolitz et al. 2021 ¹⁷	NSA	Retrospective, case series	<i>N</i> = 8	Clinical chart review	Disease course

stable vLS, and 7 patients reported worsening. Notably, in 5 patients, vLS reportedly developed, either was first diagnosed in pregnancy (reported in two studies, with onset and/or first diagnosis in 1 of 11, and 4 of 29 patients, respectively). Delivery through caesarean was attributed to vLS (in one patient owing to non-compliance) in a single patient in two studies each (1 of 11, and 1 of 29 patients, respectively). In only one patient was there a suggestion of vLS-related risk for perineal tears (1 of 9 patients, relating to 4 deliveries). One study also mentioned delayed healing of a third degree tear in one patient not under treatment (1 of 22 patients), while in another study development of vLS/koebnerization in the area of the perineal scar occurred (1 of 29 patients). Finally, worsening of vLS in the postpartum was mentioned only once with regard to three pregnancies in one woman (one study, 1 of 9 patients).

Overall, there were few studies on vLS in pregnancy. Studies were rather small, mostly retrospective and uncontrolled, rarely used standardized measures such as TCS need, and often provided little detail on treatment altogether, and therefore particularly prone to bias. Hence, no meaningful estimates can be made on the incidence of outcomes addressed. Nonetheless, the small literature base retrieved, suggests that in an apparently small proportion of patients, pregnancy and delivery may complicate the course of vLS and vice versa. Specifically, vLS was reported to possibly worsen in pregnancy, and in a few cases, to necessitate caesarean delivery, to underlie perineal trauma, and to delay or complicate perineal wound healing. While it cannot be inferred from these data whether this is also preventable, a number of complications of vLS are in fact largely prevented by long-term topical treatment.^{2,8} Furthermore, cases were described with onset either first diagnosis in pregnancy suggesting that prenatal care may provide a window of opportunity to early diagnosis and treatment of vLS.

In conclusion, while there is a defined need for prospective data on vLS in pregnancy, currently available data do suggest that in a majority of vLS patients under treatment, pregnancy does not complicate vLS and vice versa. Nonetheless, early diagnosis and proper treatment remain key, also in pregnancy, in order to possibly avoid negative outcomes relating to vLS in pregnancy. Recently published guidelines on that matter,⁹ as well as efforts to increase vulvar awareness among obstetric health care providers may further benefit women with vLS during and following pregnancy.

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CONFLICT OF INTEREST STATEMENT None to declare.

AUTHOR CONTRIBUTIONS

Britt Debaene: Data curation (supporting); formal analysis (lead); methodology (supporting); project administration (lead); validation (supporting); visualization (lead); writing—review & editing (equal). Sander Vandeweege: Data curation (supporting); formal analysis (lead); methodology (supporting); project administration (lead); validation (supporting); visualization (lead); writing—review & editing (equal). Hans Verstraelen: Conceptualization (lead); data curation (lead); methodology (lead); supervision (lead); writing—original draft (lead); writing—review & editing (lead).

DATA AVAILABILITY STATEMENT

The data that supports the findings of this study are available in the supplementary material of this article.

ETHICS STATEMENT

Not applicable.

ORCID

Britt Debaene b https://orcid.org/0000-0002-7522-2498

Sander Vandeweege b https://orcid.org/0000-0003-4776-4238

Hans Verstraelen b https://orcid.org/0000-0002-4070-1587

REFERENCES

- Lee A, Fischer G. Diagnosis and treatment of vulvar lichen sclerosus: an update for dermatologists. Am J Clin Dermatol. 2018; 19(5):695–706. https://doi.org/10.1007/s40257-018-0364-7
- Lee A, Bradford J, Fischer G. Long-term management of adult vulvar lichen sclerosus: a prospective cohort study of 507 women. JAMA Dermatol. 2015;151(10):1061–7. https://doi.org/ 10.1001/jamadermatol.2015.0643
- Vieira-Baptista P, Pérez-López FR, López-Baena MT, Stockdale CK, Preti M, Bornstein J. Risk of development of vulvar cancer in women with lichen sclerosus or lichen planus: a systematic review. J Low Genit Tract Dis. 2022;26(3):250–7. https:// doi.org/10.1097/lgt.00000000000673
- Page MJ, McKenzie JE, Bossuyt PM, Boutron I, Hoffmann TC, Mulrow CD, et al. The PRISMA 2020 statement: an updated guideline for reporting systematic reviews. BMJ. 2021;372(3): n71. https://doi.org/10.1371/journal.pmed.1003583
- The International Prospective Register of Systematic Reviews (PROSPERO). [2023 Apr 12]. Available from: https://www.crd. york.ac.uk/prospero/display_record.php?RecordID=411358
- Munn Z, Barker TH, Moola S, Tufanaru C, Stern C, McArthur A, et al. Methodological quality of case series studies: an introduction to the JBI critical appraisal tool. JBI Evid Synth. 2020; 18:2127–33.
- Moola S, Munn Z, Tufanaru C, Aromataris E, Sears K, Sfetcu R, et al. Chapter 7: Systematic reviews of etiology and risk. In: Aromataris E, Munn Z, editors. Joanna Briggs Institute Reviewer's Manual. The Joanna Briggs Institute; 2017 [2023 Apr 12]. Available form: https://reviewersmanual.joannabriggs.org/
- 8. Wijaya M, Lee G, Fischer G, Lee A. Quality of life in vulvar lichen sclerosus patients treated with Long-term topical corticosteroids.

J Low Genit Tract Dis. 2021;25(2):158–65. https://doi.org/10. 1097/lgt.00000000000599

- Kirtschig G, Kinberger M, Kreuter A, Simpson R, Günther A, van Hees C, et al. EuroGuiDerm guideline lichen sclerosus version [1]. June 2023 [2023 July 24]. Available form: https://www. guidelines.edf.one//uploads/attachments/cljzpyj9r1hqqyxjrdbpy 2szy-lichen-sclerosus-guideline.pdf
- Helm KF, Gibson LE, Muller SA. Lichen sclerosus et atrophicus in children and young adults. Pediatr Dermatol. 1991;8:97–101. https://doi.org/10.1111/j.1525-1470.1991.tb00294.x
- 11. Dalziel KL. Effect of lichen sclerosus on sexual function and parturition. J Reprod Med. 1995;40:351–4.
- Haefner HK, Pearlman MD, Barclay ML, Selvaggi SM. Lichen sclerosus in pregnancy: presentation of two cases. J Low Genit Tract Dis. 1999;3:260–3. https://doi.org/10.1046/j.1526-0976.1999. 34008.x
- Günthert AR, Faber M, Knappe G, Hellriegel S, Emons G. Early onset vulvar Lichen Sclerosus in premenopausal women and oral contraceptives. Eur J Obstet Gynecol Reprod Biol. 2008;137:56–60. https://doi.org/10.1016/j.ejogrb.2007.10. 005
- Higgins CA, Cruickshank ME. A population-based case-control study of aetiological factors associated with vulval lichen sclerosus. J Obstet Gynaecol. 2012;32:271–5. https://doi.org/10. 3109/01443615.2011.649320

- Nguyen Y, Bradford J, Fischer G. Lichen sclerosus in pregnancy: a review of 33 cases. Aust N Z J Obstet Gynaecol. 2018;58:686–9. https://doi.org/10.1111/ajo.12812
- Trokoudes D, Lewis FM. Lichen sclerosus the course during pregnancy and effect on delivery. J Eur Acad Dermatol Venereol. 2019;33:e466–e8.
- Kolitz E, Gammon L, Mauskar M. Vulvar lichen sclerosus in women of reproductive age. Proc (Bayl Univ Med Cent). 2021; 34:349–51. https://doi.org/10.1080/08998280.2021.1885093

SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.

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