1	Growing up with juvenile vulvar lichen sclerosus, the experiences and care needs of adult
2	women with lichen sclerosus since childhood: a qualitative exploration
3	Running head: Qualitative study of the experience of women with history of JVLS
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#### What is already known about this topic?

- Vulvar lichen sclerosus (VLS) may significantly impact a woman's quality of life, and needs for care and guidance are currently not being sufficiently met.
  - Although qualitative studies have recently been conducted, these focus on women with VLS in mid-life or older; less is known about the experiences of individuals with juvenile VLS (JVLS) and their preferences regarding care and guidance.

#### What does this study add?

- Through qualitative analysis of adult women who were diagnosed with VLS as juveniles (JVLS)
   the long-term impact of the disease on their lives is brought to light.
- Recommendations regarding information provision and guidance of patients with VLS as formulated in current guidelines, do not yet seem to be sufficiently implemented for women with JVLS.
- Women express a need for patient-tailored and age- and life-phase appropriate care.

#### What are the clinical implications?

• It is paramount that knowledge improves regarding (J)VLS and general vulvar health among health care professionals, especially primary care providers, and the public at large.

Individually tailored care and guidance for women with JVLS is needed, utilizing as a foundation
the determinants of therapy adherence (including, but not limited to, age, life-phase,
involvement of close ones, and understandable resources) and accounting for the influencing
factors social context and coping strategy.

#### **ABSTRACT**

- Background. Vulvar Lichen Sclerosus (VLS) is a chronic remitting condition affecting the genital skin of females of all ages. Although qualitative studies have been conducted focusing on women with VLS in mid-life or older, less is known about the experiences of individuals with VLS from childhood or adolescence onward.
- Objective. To gain understanding of the experiences of women with a history of juvenile VLS (JVLS)
  regarding the impact of the disease on their personal lives, and their experiences and needs regarding
  care and guidance.
  - Methods. A qualitative study was conducted consisting of 27 in-depth face-to-face interviews with adult women with a histologically confirmed history of JVLS, striving for maximum variation and saturation.

    Interviews were audio-taped and transcribed verbatim. A thorough thematic content analysis was performed.
  - Results. Three main themes were identified. I. Varying impact of living with JVLS: Women experienced diverse emotional and physical impact, from shame and denial to complete acceptance, from restrictions in daily functioning to no limitations. They felt hindered by their own lack of knowledge about JVLS, and generally expressed a positive influence of sharing their experiences with people close to them. II. Finding one's way in care and guidance: While navigating care and guidance, women often felt hindered by knowledge gaps among health care professionals (HCPs), lack of continuity in care and guidance, lack of life-stage adjusted and future-oriented information provision, inadequate guidance around life events,

- 1 and insufficient monitoring of determinants of therapy adherence. III. Need for patient-tailored care:
- 2 Patients stressed the need for age-appropriate and life-phase adjusted information, guidance around
- 3 life-events and compassionate contact with knowledgeable HCPs, aware of the determinants of therapy
- 4 adherence and influencing factors.
- 5 Conclusions. Age-appropriate life-phase adjusted individually tailored care for women diagnosed with
- 6 VLS in childhood or adolescence is needed. Care and guidance from childhood onward should encompass
- 7 a standard of care adapted to the individual as needs change over time. This involves taking
- 8 interpersonal differences into account, including differences in support network and coping strategies.
- 9 These findings demonstrate the need for improving awareness and knowledge about (J)VLS among HCPs,
- 10 especially primary care providers, and among the general public.

#### INTRODUCTION

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Vulvar Lichen Sclerosus (VLS) is a chronic remitting skin condition of the vulva. Diagnosis is based on clinical features, and, if inconclusive, histological examination of a vulvar skin biopsy may be diagnostic. <sup>1-5</sup> Diagnosis in juveniles is preferably solely based on clinical findings. <sup>1,6</sup> VLS may cause complaints of pain, pruritus, constipation or painful defecation, dysuria and dyspareunia and can lead to irreversible anatomical changes of the vulva. <sup>2,7</sup> VLS in adults also increases the risk of developing vulvar squamous cell carcinoma. <sup>8</sup> The aetiology of VLS is not fully understood, but an association with autoimmune disease and a genetic predisposition are recognized. <sup>2,6,9-11</sup> Though not curable, the symptoms of VLS are treatable and progression of disease can be controlled with regular application of steroid ointments or calcineurin inhibitors combined with emollients. <sup>3,6,12</sup> The prevalence of VLS in girls is estimated to be at least 1:900. <sup>13</sup> However, this number is likely to be an underestimation. <sup>2,7,14</sup> Though VLS can manifest itself at any age, it is often stated that it develops most often in prepubertal or postmenopausal

females.<sup>4,6</sup> We speak of juvenile vulvar lichen sclerosus (JVLS) when the disease occurs in childhood or adolescence.<sup>15</sup> Despite the fact that research on VLS and JVLS has increased in recent years, major knowledge gaps remain.<sup>15,16</sup>

The burden of having a chronic vulval skin condition such as VLS has a negative impact on quality of life (QoL).<sup>6,14,17-20</sup> Several qualitative studies have been conducted to gain a better understanding of the experiences of women with VLS, though focusing on women diagnosed as adults, showing that the needs of women with VLS are currently not sufficiently met by the healthcare system.<sup>7,21-26</sup> Little is known about the experiences and impact on the lives of children or adolescents with JVLS and their caregivers nor the repercussions in adulthood, including the (sexual) development and obstetric experiences which will mostly take place many years after the initial diagnosis of JVLS.<sup>15</sup>

The aim of this study is to gain understanding of the experiences of women with a history of juvenile VLS (JVLS), regarding the impact of the disease on their personal lives, and their experiences and needs regarding care and guidance during childhood, adolescence and adulthood.

## **MATERIAL AND METHODS**

#### Study design

- A qualitative study was designed to gain an in-depth understanding of what females with a history of

  JVLS experience from time of diagnosis in childhood onward. Individual interviews were chosen over

  focus groups, as they are considered most appropriate for exploring sensitive topics.<sup>27</sup>
- 20 Reporting followed the Standards for Reporting Qualitative Research. <sup>28</sup> The Institutional Review Board of 21 Erasmus MC approved the study (MEC-2019-0474, amendment A-0004 NL63335.078.19 v11).

#### Selection and recruitment of participants

2 The study group was recruited from the group of adult women with histologically confirmed JVLS in the

Netherlands who were aged 18 years or younger at time of diagnosis and fluent in the Dutch language

and who had participated in a previous study. 1,20 All eligible women (n=79) were sent an information

folder by email and post and invited to participate, aiming for maximum variation. Twenty-nine women

gave their written informed consent, and interviews were completed with 27 women. Travel expenses

were reimbursed.

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#### Data collection

The participants visited the out-patient department of the Erasmus University Medical Center from June to October 2022. A clinical examination of the vulva was performed simultaneously by three experienced vulvar clinicians (CLMvH or KMGJW, BM and MJtK-B). A clinical assessment of disease severity was made and shared with the participant who was given the opportunity to ask questions. An audio-recorded interview was then conducted in Dutch (BM, MJtK-B). If the participant wished, an accompanying family member joined the interview. Duration of the interviews ranged from 15-55 minutes. An interview guide (Supplement 1) was followed. Investigator triangulation was used to reduce bias for both data collection and data analysis by having two researchers perform and analyse the interviews. The patient characteristics were collected and stored using the LIMESURVEY and GEMSTRACKER software.

#### Data processing and analysis

The audio-recordings of the 27 interviews were transcribed verbatim and anonymised. Age at time of biopsy, current age, ethnic background and education were categorised to ensure anonymity. The transcripts were analysed using the qualitative data analysis software NVivo version 12 (QRS

<sup>&</sup>lt;sup>a</sup> Reporting of physical findings <paper accepted and being processed for publication in the Journal of Lower Genital Tract Disease>

International). A thorough thematic content analysis, based on the grounded theory method, was performed by developing an analytical coding framework and applying it to interpret the data. <sup>27,30-33</sup>

Firstly, the transcripts were summarised in order to become familiarised with the data (BM, HJK).<sup>33</sup> Then eight transcripts were openly coded by one researcher (HJK) and checked and complemented by a second researcher (BM). Disparities in coding were resolved through discussion (HJK, BM). After consensus was reached, the list of open codes was organised into potential categories and subcategories. Using this coding scheme, the remaining interviews were axially coded and the coding scheme was refined and complemented. The final coding scheme (Supplement 2) was checked and approved by a clinician-researcher (MJtK-B) and a psychologist (ML). Thematic saturation was reached after 20 interviews, i.e. no new relevant (sub)themes emerged from additional data. Nevertheless, all interviews were analysed, given that these had already taken place and could possibly and in fact did reveal subtle differences.

Then, selective coding took place in which the final subthemes and main themes were defined and named and a thematic network was constructed which was presented to the team for discussion. Through analysis of the thematic network a conceptual framework was constructed. Again, these results were presented to the multidisciplinary research team for discussion and refinement, resulting in consensus on the interpretation.

#### Research team and reflexivity

The researchers BM (MD, IFEPAG-fellow) and MJtK-B (MD, PhD) are gynaecologists with decades-long experience in vulvar disease and paediatric gynaecology. HJK (MD) participated in this project during her masters in medicine. IAMvdA (MD, MSc, PhD, gynaecologist), CLMvH (MD, dermatologist), KMGJW (MD, dermatology resident), and CWB (MD, PhD, gynaecologic oncologist) are clinicians experienced in anogenital disease. SGMAP (MD, PhD, paediatric dermatologist-immunologist)

1 is an expert clinician-researcher regarding children with dermatological diseases. ML (PhD, psychologist)

2 is an experienced qualitative researcher. By creating a multidisciplinary team, different viewpoints were

brought to light. In addition, throughout the process of data analysis, a number of meetings took place

for input and approval of the interpretation of the data. All researchers reflected on their role within the

research prior to analysis, and all members of the team have an affinity with the concerns of patients

with VLS and JVLS.

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#### RESULTS

#### Patient Characteristics

10 The characteristics of the 27 participants are shown in Table 1 and categorized in Table 2. Median age

was nine years at biopsy and 29 years at the interview. In a recent publication quantitative data on

quality of life and vulva specific questionnaires completed by these 27 participants was presented.<sup>b</sup>

#### Main themes and influencing factors

Three main themes were identified: I. Varying impact of living with JVLS; II. Finding one's way in care and

guidance, i.e. navigating the patient journey, and III. Need for patient-tailored care and guidance. Each of

these themes includes subthemes as outlined Table 3. Two major influencing factors were recognised: A.

Coping strategy and B. Social context. These themes and influencing factors, visualized in a thematic

network (Figure 1), act as barriers or facilitators towards each other. The result of the analysis of the

dynamics of the interaction of the themes and influencing factors, including the different phases of life in

<sup>&</sup>lt;sup>b</sup> Reporting of answers to questionnaires <paper accepted and being processed for publication in the Journal of Lower Genital Tract Disease>

- 1 which the subject may find themselves, each phase encompassing its own unique circumstances, is
- 2 conceptualized in Figure 2.

#### I. Varying impact of living with JVLS

- 4 The experienced impact of living with JVLS varied among the interviewees. Differences in experiences
- 5 were closely related to the woman's severity of complaints, her coping mechanisms, caregiver
- 6 involvement, and her own understanding of VLS. Table 4 shows illustrative quotes for this theme. Four
- 7 subthemes were identified.
- 8 i. Impact on women's personal lives and daily activities ranged from extensive limitations and difficulties
- 9 accepting their VLS to none at all. For some the disease had become a part of who they are. Differences
- 10 in coping influenced any experienced or perceived obstacles. During daily activities that could cause or
- increase complaints, such as cycling or wearing underwear or tight clothing, women were reminded of
- 12 the fragility of their vulvar skin. Urination, the use of menstrual products and matters of sexuality posed
- 13 (mental) barriers for some.
- 14 ii. Emotional impact of receiving the diagnosis as a juvenile. Some felt anger or frustration for being
- different from their peers. Others tended to downplay or deny their complaints. Feelings of loneliness
- 16 and insecurity were common; feelings of shame and taboo contributed to delays in seeking care. Some
- 17 blamed themselves for their complaints. Worries such as recurrences of disease, the increased risk of
- 18 cancer, potential anatomical changes, and the possibility of vaginal childbirth also had an emotional
- 19 impact.
- 20 III. Feeling insufficiently equipped with knowledge about (J)VLS, often led to a sense of isolation among
- 21 the women. Not being knowledgeable about normal vulvar anatomy, even up to the present, sometimes
- 22 led to self-blame. Women described that their parents or guardians did not always understand the

- 1 information given by the health care professional (HCP), corroborated by several mothers who joined the
- 2 interviews.
- 3 iv. Effect of sharing experiences with others. Women who were able to talk about their condition with
- 4 their caregivers, friends or partners often felt supported and understood. For others shame and taboo
- 5 formed a barrier to discuss their condition. Contact with fellow sufferers had a positive influence for
  - some, while for others their worries were augmented by information and horrific stories shared on social
- 7 media.

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#### II. Finding one's way in care and guidance, i.e. navigating the patient journey

- 9 While navigating care and guidance, women often felt hindered by several barriers as described in the
- 10 five identified subthemes, illustrated by the quotes in Table 5.
- 11 i. Knowledge gaps among HCPs. The participants felt that knowledge gaps of HCPs led to delays in
- diagnosis and care. This also led to feelings of being misunderstood and inadequate guidance around life
- 13 events, e.g. being cared for by a midwife who is completely uninformed about VLS. Whether or not the
- 14 HCP was knowledgeable influenced how women perceived their disease, and some expressed
- sometimes feeling misunderstood. Furthermore, women explained that the physician did not always
- 16 check if the child and her parents understood the information provided.
- 17 ii. Lack of continuity in ongoing (follow-up) care was often reported and perceived as a shortcoming.
- 18 Sometimes the patient was told by the HCP that follow-up was no longer necessary (contrary to current
- 19 guidelines). Participants described that their caregiver or they themselves ended the contact because of
- 20 the absence of complaints, loss of interest during puberty or adolescence, or not realising that
- 21 continuing care was advised.
- 22 iii. Lack of life-stage adjusted and future-oriented information provision at different stages of growing up
- 23 was experienced as a shortcoming by some women, while others were content with the information

given. Several women reported that new questions arose as they grew older while, due to lack of 1 2 continuity, they were not able to ventilate their questions. For example, it was noted that the possibility 3 of JVLS affecting their (future) sexual activity was not always discussed during follow-up consultations, 4 even when the juvenile was approaching an age when she might want to explore her sexuality. 5 iv. Inadequate guidance around life-events was experienced by women. In particular, interviewees who 6 had given birth described missing adequate guidance during pregnancy and labour which resulted in 7 being severely anxious about giving birth. A number of women described that the pain felt during labour brought back traumatic memories of their childhood vulvar complaints. Conversely, women with a 8 9 knowledgeable and empathic gynaecologist or midwife, felt supported and reassured. Consultations, as described by the participants were, in general, focused on practical aspects of disease 10 11 control with little room for questions. Some were content with not having talked with their physician 12 about sex as an adolescent, preferring to avoid the topic of the potential influence of VLS on sexuality, while others had missed having an opportunity to discuss this topic with their physician. 13 14 v. Insufficient monitoring of determinants of therapy adherence. According to interviewees, various factors influenced therapy adherence: absence or presence of complaints; experiences with ointments or 15 16 creams; regular surveillance by an HCP; entering a new phase of life; involvement of parents (social 17 context); understanding by patient and caregivers of the implications of VLS; and understanding the protective effect of therapy for long-term consequences. 18

## III. Need for patient-tailored care involving appropriate and compassionate care and guidance.

The need for patient-tailored care and guidance, which involves appropriate information provision and compassionate care and guidance, consisted of two subthemes with illustrative quotes being shown in Table 6.

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1 i. Age-appropriate and life-phase adjusted information provision and knowledge at all levels of society.

Participants indicated that information provision for young children should mainly focus on the present,

with attention to practical tips regarding therapy adherence, ointment schedules and possible

hindrances, for example tight clothing or underwear. It was noted that, for children the chronic aspect of

JVLS should be addressed, and motivating the patient to be compliant without being ominous.

As the child grows older, further explanation, also focusing on sexuality and psychological impact, should be offered at appropriate moments in time during regular follow-up consultations, taking the health literacy and language proficiency of the juvenile and their caregivers into account. Suggestions include hand-outs, preferably in the native language of the caregiver or an informative video, discussing the availability of contact with fellow sufferers via social media or other channels. Participants expressed the desirability that the HCP keep the patient abreast of new insights on all aspects of VLS or JVLS that emerge, including but not limited to therapy options.

Participants discussed deficiencies in their own knowledge and that of the general public regarding vulvar anatomy and disease which led to a higher threshold to reveal their complaints. More information in high school biology lessons, as well as more accessible information online and in popular magazines was proposed.

ii. Patient-tailored and compassionate care and guidance based on a standard of care. The participants

*ii. Patient-tailored and compassionate care and guidance* based on a standard of care. The participants stressed each individual's unique situation. Some juveniles can rely more on their social network than others, due to differences in health literacy and involvement of caregivers. Receiving compassionate and empathetic care by the HCPs was perceived as beneficial and important for how one experiences the disease. The women also discussed that as juveniles with VLS go through different stages of life different needs for care and guidance may arise as they mature, and these needs should be met in an age-appropriate and life-phase adjusted manner and with empathy and compassion.

#### **DISCUSSION**

We report on a qualitative study of adult women who were diagnosed with VLS during childhood or adolescence. The analysis identified three major themes, namely, varying impact of living with (J)VLS, navigating the patient journey with care and guidance, and, finally, the need for patient-tailored care involving appropriate and compassionate care and guidance. The experiences of the child, adolescent

and adult with JVLS are influenced by their coping strategies and their social context.

#### Varying impact of disease

The present study shows that while the experience of these women in many aspects concurs with adult onset VLS regarding the impact of the disease, there are also some differences. In previous qualitative research on women diagnosed with VLS at an adult age, the participants were often recruited through social media and special interest groups, and often older. Common themes in those studies were: dealing with the chronic aspect of VLS, feelings of taboo and shame and the consequent feelings of loneliness and isolation, and disruption of sexual functioning. 7,21,22,24,26,34 In addition to those themes, our young participants (median age 29 years) often focussed on dealing with the disease in daily life and their own (lack of) knowledge about VLS. Coping strategies had been developed at an early age, ranging from denial to acceptance or even incorporating the VLS into their own sense of being, as had been described in children with severe chronic skin disease. 35,36 De Maeseneer et al. performed a study among children with severe chronic skin disease, and ascribed this phenomenon to the so-called 'disability paradox' of Albrecht and Devlieger. 35,36 This might also explain the gap between the perceived quality of life and the observed severity of VLS activity among our study group. 6

<sup>&</sup>lt;sup>c</sup> Same reference as in footnote a and b.

#### Navigating the patient journey

Arnold et al. showed that the need for contact with fellow sufferers varied. <sup>21</sup> The participants in our study were not likely to be active on online forums. Though they expressed a positive attitude toward the existence of social media groups, they felt little affinity with these groups.

A child is more likely to be dependent on their support network than an adult may be. The degree to which parents understand the child's illness and care influences treatment adherence and whether or not they will remain in care. Feelings of loneliness, shame or worries are related to the extent to which complaints can be openly discussed and whether questions can be asked or answered by caregivers. In this study there was great diversity in the support networks, including foster parents, single-parent families, and sometimes parents with a substantial language barrier. Our findings regarding the needs and preferences for care and guidance underscore and expand on the needs identified in previous qualitative studies and established guidelines. 3,5,21,22,24

Knowledge gaps among HCPs, with delay of diagnosis, have previously often been described, and the present study substantiates that finding. <sup>21,22,24</sup> The results of the present study also highlight the fact that recommendations from current guidelines have not yet been adequately implemented in daily practice for children and young women diagnosed with VLS, especially with regard to maintenance treatment, <sup>12</sup> follow-up, actively enquiring about difficulties in daily life, and including age-appropriate life-phase adjusted questions. This may reflect knowledge gaps among HCPs or it could be a consequence of the fact that many women in our study had been diagnosed decades ago, before this knowledge was available. This fact demonstrates the need for HCPs to stay abreast of new developments.

#### Need for patient-tailored and compassionate care

An important theme in the current study is that following a diagnosis of JVLS, patients should be offered information repeatedly throughout their lives in an age-appropriate manner. Due to lack of

ongoing care, life phase-adjusted and future-oriented information provision regarding e.g. sexuality, giving birth or the climacteric may be lacking. Regular follow-up consultations appear to strengthen therapy compliance; the ongoing conversation with the doctor motivates compliance and being alert to changes in one's complaints or vulvar anatomy.

Common JVLS care, as perceived by many of our participants, gives limited attention to the determinants of therapy adherence. Adherence to therapy has been shown to be critical regarding prognosis of permanent vulvar architectural damage in girls with JVLS.<sup>37</sup> Taking the determinants of therapy adherence including the individual's coping strategy and their social network into account during consultation, would result in more patient-tailored care. Supplementary digital or written information was advised, but is not a substitute for actively asking about the patient's needs and showing compassion in care and guidance. Beyond routine check-ups, planning a more extensive consultation when approaching each new phase of life may create a sense of being heard. This entails, for example, being given enough opportunity to ask questions, having a dialogue during consultation, the HCP broaching sensitive topics like sexuality in an open manner. Participants reported the positive effect of having a say in the choice of by whom and where follow-up takes place, giving a sense of self-control.

The aforementioned study of De Maeseneer et al. concluded that children with a severe chronic skin disease were in need of 'a more personalized integrative multidisciplinary and systematic transmural approach covering all aspects of life during their lifetime'. The current study is in support of this viewpoint and shows that this is applicable to children and adolescents with VLS.

#### Recommendations

For professionals seeing patients with VLS and JVLS there should be adequate time and capacity to address the relevant aspects of care and guidance. Table 7 has been formulated to assist the physician during consultations, showing recommended topics that should at least be touched on at each consultation, not just at initiation of care. We believe that even with limited resources the physician can

give compassionate professional care adjusted to the needs of the patient over time. The findings of our exploratory study could be used as a basis for a prospective quantitative study assessing the impact of living with JVLS among both patients and caregivers from diagnosis onwards in a larger and more diverse population. This could lead to development of a standard of care with a guide for the contact between a patient, the caregivers and the HCP, creating a basis for improving clinical practice.

Strengths of this study include the fact that all participants had biopsy confirmed disease and were sought out by us, ensuring reliable inclusion despite the length of time since diagnosis. Most other studies included participants found through online platforms which may have yielded a bias to those with more severe disease or misdiagnoses. The retrospective aspect of this study allows for a long-term analysis and exploration of the experience and needs of care over time and in different phases of life. As thematic saturation was reached and the patient characteristics varied on most topics thought to be relevant, we deemed the sample size to be sufficient and assume that the identified themes are transferable to similar settings. Moreover, the reporting of this study took place according to the Standards for Reporting Qualitative Research, facilitating transparency and further enhancing transferability.<sup>28</sup> Finally, the thorough thematic content analysis, based on the grounded theory method, contributes to robust and reliable results.<sup>30-33</sup>

Limitations include the following. Observer bias cannot be completely ruled out, though this is reduced by investigator triangulation, with multiple researchers involved in both data collection and data analysis as well as reflexivity of the research team. <sup>29,38</sup> The retrospective exploration many years after initial diagnosis might have led to lapses in memory or recall bias. Some women were not even aware of the childhood diagnosis although they did recall that something had taken place which they experienced as traumatic without knowing the details. Variation in age, ethnic identity and education level was not extensive. The perspective of the caregiver of a child with JVLS goes beyond the scope of the current

- 1 study but is an important aspect and should considered in future research to fully understanding the
- 2 experience of the patient with JVLS.

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#### CONCLUSION

While expert clinicians well-versed in the care and guidance of patients with JVLS and VLS may consider the findings from the current study so fundamental that they hardly need mentioning, the participants' experiences show there are still significant gains to be made in the care for these girls and women. Much could be gained by improving the information that is shared with general health care professionals at all levels including those not specialized in vulvar health. Although the current guidelines of the British Association of Dermatologists do not specifically discuss age-appropriate counselling, those guidelines do recommend that well-informed professionals should be seeing these patients. 5 In a future revision of the guidelines a recommendation for an age-appropriate and patient-tailored approach could be incorporated. In the meantime, as a starting point, awareness of existing expert-based guidelines should be improved within the larger community of professionals seeing children and adults with LS. This study demonstrates the need for patient-tailored care following a diagnosis of JVLS, taking the determinants of therapy adherence and the influencing factors social context and coping strategy into consideration. A standard of care should be aspired to, with a patient-tailored approach and with appropriately timed extra consultations for counselling at each new phase of life. Working together, professionals and patient organisations could play an important role in informing and educating the general public about (J)VLS. Knowledgeable HCPs are a prerequisite to reducing the inadequacies of care and guidance that so many patients with JVLS and VLS have experienced. As juveniles with VLS go through different phases of life, different needs for care and guidance may arise as they mature, and their needs should be met with empathy and compassion in an age-appropriate and life-phase adjusted manner.

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#### Figure legends

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- Figure 1. Thematic network of main- and subthemes and influencing factors regarding the experiences of
- women with a history of JVLS and their needs for care and guidance
- 36 The interrelationships between the (sub)themes are indicated by connecting lines.
- 38 Figure 2. Schematic overview themes: Experiences of women with a history of JVLS and their needs for
- 39 care and guidance.
- 40 The three phases of life (left, grey inner arrows), the two main themes (left, dark blue outer arrows), the
- 41 two influencing factors (left, grey outer circle), resulting in the third, and overriding main theme (right,
- 42 grey inner circle).

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Table 1. Individual participant characteristics (n=27)

				11-27)			
Participant'	Age at	Age at	Ethnic/	Educational status	Sexual	Having ever	Status of VLS
s number in	time of	time of	cultural		identity	given birth	at time of
quotes	interview	biopsy	identity				interview*
	(years)	(years)	,				
1	30-39	2-5	Indonesian	Post-secondary	Heterosexual	Yes	Moderate
-	30 33	23	Dutch	vocational education	ricterosexaar	165	Wioderate
2	20-29	6-12	Indonesian	Post-secondary	Not reported	No	Not active
2	20 23	0 12	Dutch	vocational education	Not reported	NO	Not active
3	<20	6-12	Dutch	Higher professional education	Heterosexual	No	Not active
4	30-39	13-18	Dutch	Post-secondary vocational education	Heterosexual	Yes	Moderate
5	20-29	2-5	Dutch	Higher professional education	Not reported	No	Mild
6	20-29	6-12	Dutch	Post-secondary vocational education	Homosexual	No	Moderate
7	20-29	6-12	Dutch	University degree	Heterosexual	No	Mild
8	20-29	6-12	Dutch	University degree	Heterosexual	No	Mild
9	20-29	6-12	Turkish Dutch	Post-secondary vocational education	Heterosexual	No	Mild
10	30-39	13-18	Dutch	Post-secondary vocational education	Heterosexual	Yes	No evident LS
11	20-29	6-12	Turkish Dutch	University degree	Heterosexual	No	Not active
12	20-29	13-18	Dutch	Post-secondary vocational education	Heterosexual	No	Not active
13	30-39	6-12	Dutch	Post-secondary vocational education	Heterosexual	Yes	Not active
14	30-39	6-12	Dutch	Higher professional education	Heterosexual	No	Not active
15	<20	2-5	Dutch	Higher professional education	Heterosexual	No	Moderate
16	20-29	13-18	Dutch	Post-secondary vocational education	Heterosexual	Yes	Mild
17	20-29	13-18	Dutch	University degree	Heterosexual	No	No evident LS
18	20-29	6-12	Dutch	University degree	Not reported	No	Mild
19	40-49	13-18	Dutch	University degree	Heterosexual	Yes	Severe
20	30-39	2-5	Dutch	Higher professional education	Heterosexual	Yes	Not active
21	20-29	2-5	Mixed Ethnic Background	Higher professional education	Bisexual	Not reported	Severe
22	20-29	6-12	Dutch	Higher professional education	Heterosexual	No	Moderate
23	30-39	13-18	Dutch	Secondary education	Heterosexual	No	Moderate
24	20-29	13-18	Dutch	Post-secondary vocational education	Homosexual	No	Mild
25	30-39	2-5	Dutch	Higher professional education	Heterosexual	Yes	Moderate
26	20-29	13-18	Dutch	Higher professional education	Heterosexual	No	Mild

27	30-39	6-12	Dutch	Higher professional	Heterosexual	Yes	Not active
				education			

<sup>\*</sup>Based on physical examination prior to interview.

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# 3 Table 2. Participant characteristics, adult women with JVLS (n=27)

PARTICIPANT CHARACTERISTICS	N
Age at time of biopsy (years)	
2-5 (preschool child)	6
6-12 (child)	12
13-18 (adolescent)	9
Age at time of interview (years)	
20-29	15
30-39	10
40-49	2
Ethnic/cultural identity	
Dutch	22
Non-Dutch or mixed Ethnic Background	5
Educational status	
University degree	7
Higher professional education	9
Post-secondary vocational education	9
Secondary education	2
Sexual identity	
Heterosexual	21
Homosexual	2
Bisexual	1
Not reported	3
Obstetric status	
Never pregnant	17
Ever pregnant	10
Status VLS at time of interview*	
Active Mild	0
	8
Moderate	7
Severe	2 8
Non-active with residual signs	2
No evident LS	2

<sup>\*</sup>Based on physical examination prior to interview.

Abbreviations: JVLS juvenile vulvar lichen sclerosus; VLS vulvar lichen sclerosus

Table 3. Outline of themes in care and guidance of women with Juvenile Vulvar Lichen Sclerosus

	-
I. V	arying impact of living with JVLS
i.	Impact on women's personal lives and daily activities
ii	. Emotional impact of receiving the diagnosis as a juvenile
ii	i. Feeling insufficiently equipped with knowledge about (J)VLS
i۱	v. Effect of sharing experiences with others
II. Fi	nding one's way in care and guidance, i.e. navigating the patient journey
i.	Knowledge gaps among health care professionals
ii	. Lack of continuity in ongoing (follow-up) care
ii	i. Lack of life-stage adjusted and future-oriented information provision
i۱	. Inadequate guidance around life-events
V.	Insufficient monitoring of determinants of therapy adherence
III. N	eed for patient-tailored care involving appropriate and compassionate care and guidance
i.	Age-appropriate and life-phase adjusted information provision and knowledge at all levels of
socie	ety
ii	. Patient-tailored and compassionate care and guidance

# 2 Table 4. Theme: Varying impact of living with JVLS

Subthemes	Quotes
Impact on wome	n's personal lives and daily activities
Complaints in	'I can't remember much, only that there was prickly and burning sensation and that I
childhood and	avoided bowel movements. In the evenings I asked to sit in the bathtub and the burning
adolescence	and prickly feeling would go away after that my mother put ointment on. I usually slept
	without knickers but I scratched it completely open because of the itching.'
Difficulties in	'For a long time all went well. But when I started having my periods things went wrong,
daily life	sanitary towels didn't work, I was always wet; but by trying different towels and changing
	often I found my way. Sometimes I had to use 8 towels a day. It was expensive, I even had
	to change pads at night.'
	X) Y
	'The only thing I can think of is, for example, with cycling, that was the only problem, that
	I often had complaints when on my bicycle, and at a certain point I got a special rounded
	shaped saddle meant for wearing a skirt.'
Emotional impac	t of receiving diagnosis as a juvenile: (from shame and denial to complete acceptance)
Feeling normal	'As I child, you want to feel you are normal and when you're lying there, you really feel
vs. different	like this is not normal. Other kids my age didn't have to go through that. So that's why it
	was a trauma every time, even before I had to go to the appointment itself. At a certain
	point my mother didn't tell me that we were on the way to the hospital.'
Y.	
	'In high school I became aware in biology lesson. We had pictures in the textbook and
	then I saw that it looked different to mine. So I thought "oh yeah, I'm sick".'
	'As far as I know, it all looked normal, no two are the same. So, I never thought "blimey, it
	looks strange" or that I felt less or more than another (because of it).'
Self-blame	'I thought that I had caused it myself, that I shouldn't scratchI thought it was my own

# fault. Maybe if I had known that there could be something wrong I would have gone to the doctor sooner.' Dealing with 'Moisturising is important, but otherwise....just take it as it is, just moisturise and accept. the diagnosis Being dramatic is just wasted time.' 'In itself, sexual intercourse is fine, only, if you touch the clitoris with a finger, um, I don't know, it's just not really a nice feeling for me. It feels like a wound being rubbed. Yes, that, and that used to be different of course. So yes, I notice a difference. I hardly have any problems, maybe four times a year.' 'I think I didn't even tell my friends, I can't remember, I think that I felt alone....and that I went through it on my own. That's weird, that you are all alone your whole life. Then you're your own discussion partner, your own psychologist, your own coach. So that's in your own head, from 5 years old on. (Interviewee becomes emotional).' 'When it (the LS) returned when I was 12 or 13 years old I completely panicked.' 'Straight away I thought I wouldn't be able to have children and that I would gradually close-up completely. My sister said "I'll be a surrogate mother for you." That's how far I was thinking.. although at that time I wasn't even thinking about having children yet. Now I realise, blimey, I could have saved myself a lot of sadness and fear if I had known how it really was, something like that." Feeling insufficiently equipped with knowledge about (J)VLS 'I still don't dare to go to the toilet anywhere. I can't. I think to myself, "maybe it's dirty, or I'll catch something, an infection." I'm afraid of a bladder infection. So I've had days, days when I've held my urine up for a very long time.' 'Yes, at the beginning of my pregnancy I didn't want to apply ointment because I wanted to avoid harming the baby. But at a certain point the itching was so bad that I just had to apply ointment to calm it down.' Effect of sharing experiences with others When I was younger I found it more difficult, but now I live in a house with four female roommates, they know about it and I can talk to them about it. I could talk to my mother about it, she understood and that was perfect.'

'I knew that I had it, I didn't want to know, didn't wat to have it, and I didn't talk about it to anyone. In puberty my interests were not at all there. I'd rather not have it and acted

'I visited the Facebook group online, but those are such scary stories, I think, "Is this what I want?" Because I'm young maybe progression is slower, so that I'm lucky. So, yeah, do I

1

as if it didn't exist.'

really want to know everything that's awaiting me?'

# 1 Table 5. Theme Finding one's way in care and guidance, navigating the patient journey

Subthemes	Quotes
	of health care professionals
3 3 1	'There, in the local hospital, they didn't know what it was, I went there two or three times, and I myself had to ask for a referral to an academic hospital.'
	'She [the GP] had inserted an IUD twice in the course of time, so if there was something there that I didn't see she must have seen it, I assumed. She said, "It's a yeast infection, so here's some medicine, and it will get better".'
Lack of continuity	y in ongoing (follow-up) care by health care professionals
	'I was on my own as a teenager. No [professional] saw me; therefore, I had to look everything up myself. And then at some point they advised an operation to open it up (before I even ever had sex). After the operation I was left on my own again.'
	'Yes, my mother said that I should go for a check-up twice a year. But because I wasn't having any problems, no symptoms, we didn't go back. So actually in the period between 9-10 until 25 years old I'd only had a check-up twice.'
Lack of life-stage	adjusted and future oriented information provision
	'When you ask which information I'd like to receive, then I'd like to have the information about "now". So when I was 20 I'd didn't want to talk about giving birth yet, and now I'm close to 40 and would like to hear about the menopause but that hasn't happened yet.'
	'I think I was too young then for more information. But at some point that should have been given. Because I always went back just for check-ups, and I actually learned more through Google and self-examination than having received information.'
Inadequate guida	ance around life events
	'I think, especially when you approach puberty and when you become sexually active, at those moments, you have to know about the importance of applying ointments and checking that it looks alright yes, that you're told "pay special attention to this".'
	'During my first pregnancy I'd been to the gynaecologist, the midwife had referred me to see if it was possible to plan for a normal vaginal delivery. She (the gynaecologist) believed it was possible, so I wasn't apprehensive about it.'
	'So, I let my midwife know that I was stressed about giving birth. We had a delivery-plan but I thought "that doesn't ease my worries", something like that. I was given a kind of neutral answer to all my questions. Most of my symptoms came after the delivery. But I was scared to death of the delivery, really scared. I thought "I'll tear completely open, everything will be damaged and it will not heal." So the delivery was a drama.'
Insufficient moni	itoring of determinants of therapy adherence
Absence or	'At the moment I'm not thinking about it daily, only if I happen to have symptoms, then
presence of complaints	I think, "Oh, yes", and I apply some ointment once in a while. But if I don't have symptoms then I just forget, like you do. And if you have check-ups you have a rhythm, of course.'
Experiences with ointments and creams	'That ritual of taking a shower and then applying ointment, works pretty well for me. I'm doing better than I did in the past. I had been prescribed one type of ointment or cream that I didn't like, which was the reason why I didn't use anything.'
	'That it's something that always is part of me, so it's something I like to ignore. And as

	long as I use the ointment almost every morning, then I think for that moment, "oh
	dear", and I find it nice that I can ignore it the otherwise.'
Regular follow-	'And I don't think I had any more check-ups. So that makes it easy be less attentive, so
ир	each year I think, "no, not this year". Just like I'm thinking right now, "o.k., maybe I
consultations	should go." Somehow I think that a bit of pressure helps.'
Entering a new	'I think I did have a phase when I was more negligent about applying ointments, and
phase of life	when you're young you don't have all that many ideas about it.'
Involvement	'Your background plays a big role in that. To what extent did your parents include you
caregivers	in what was going on. My parents were given an ointment that we had to keep
(social context)	applying, only there was some miscommunication. My mother thought that we just had
	to finish the one tube, but the doctor meant life-long. She feels guilty about both these
	things, about not knowing enough, but especially that she did not realise she had to
	continue with the applications.'
Understanding	'My ointment is lying there, but I don't apply it too often because I think, the less
of VLS	sometimes, the better. Then, anyway, that's what I thought. So a tube of ointment
information by	would last maybe 2 to 3 years.'
patient and	'I know that I should use the ointment, but with me, well it's just that I'm afraid of the
caregivers	Dermovate, you know, that hormone ointment, that if I use it I'll get wounds. I'm so
	afraid of that happening.'
Protective	'I did start with ointment application, not so much for the present, but I use
effect long-term	maintenance for prevention for when I'm older.'
consequences	

# Table 6. Theme: Needs for patient-tailored care involving appropriate and compassionate care and guidance

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Subthemes	Quotes
Age-appropriate	and life-phase adjusted information provision and knowledge at all levels of society
	'Maybe it would be good to speak to the adolescent alone. Because a teenager doesn't really want to discuss everything with their parents. At least I didn't want to. Maybe some people do.'  'What really helped me was that I looked on the internet. One of the first things I saw was an 'influencer', who had also been diagnosed and she had a lot of female followers, maybe between 18 and 28 years old In her video she talked frankly about what her symptoms were and that the GP thought it was a yeast infection. When I read that I recognized this, and all the posts of young women who had the same experience. So that really helped me and I also found it refreshing. Because all the information that is available and all the research is in such difficult medical language.'
<b>&gt;</b>	'Yes, I'm sure that information in my native language would have helped. Even if the name of the disease is the same, if you are given a brochure or written flyer that's in your own language you can absorb it better, rather than superficially skimming through it.'
	'Though I know one might think that a 13 year-old and 16 year-old are of similar ages, the 13 year-old is more likely to think, "it doesn't matter much if I don't use the ointment sometimes", while as a 16 year-old you're more matureso the discussion

that one has with their doctor at 13 should be repeated at 16. I don't know if that is attainable, but the discussion should be repeated, say, every four years or so.'

'Actually, I was usually only asked how many symptoms I had, how often I applied ointment, if I had any pain or complaints with daily living, or if I had any difficulties. But I think that from a certain age onwards maybe then you should be asked more directly about sex partners. Not much was ever asked about that. They asked if I was sexually active, but then didn't ask any further.'

'That would have been better if someone had told me, "That seam in your underwear may be unpleasant". For trousers they did tell me that if they're too tight it would rub and be uncomfortable. It would have been better if someone had taken the trouble to tell give me advice in the choice of underwear, something like that.'

'I do think that for children it's better not to make an issue of it, just name it and move on ... In daily life I didn't have many problems with it, so I think, don't make it a bigger problem than it is.'

'Through this questionnaire I learned that there is platform for LS, I really didn't know about that. So I think that it is great that there's this platform, and maybe I didn't know about it because I've been going to the doctor for so long, nobody thought to tell me.'

# General education

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2 3 4 'But maybe they should put information about it in magazines for parents, a few that are popular, with information about if you child is scratching their bottom, what you should know. Websites for parents with questions about their kids are full of information, they lead you to the websites of those magazines.'

#### Patient-tailored and compassionate care and guidance

'What I remember is that when my gynaecologist told me I could go to the GP for my check-ups I told her that I didn't think that was really a good idea. I felt that a GP has much less understanding because they rarely see it. So I chose to continue having my check-ups with the gynaecologist.'

'Nobody ever asked before now, "How do you feel about this?" No, they never asked that. I think that for children the psychological aspects are important. But nobody looked at that.'

'As a child, and in the years that followed I had the same gynaecologist every visit, and then you get to know each other. So I didn't mind it so much, and (when I was older) I just went on my own without my parents, of course.'

'Since last year I visit a gynaecologist that, personally, I prefer. Maybe because they're new, and want to get to know me. She asked much more thoroughly including how it was for me [to have the LS] and that was much more pleasant than previous experiences.'

## 1 Table 7. Points of attention during every consultation with a patient with JVLS

# Social context

#### **Coping strategies**

# Determinants of therapy compliance

- Absence or presence of complaints
- Experiences with ointments and creams
- Regular follow-up consultations
- Entering a new phase of life
- Involvement caregivers (social context)
- Understanding of VLS information by patient and caregivers
- Protective effect long-term consequences

# Relevant personal questions the patient has

**New insights** 

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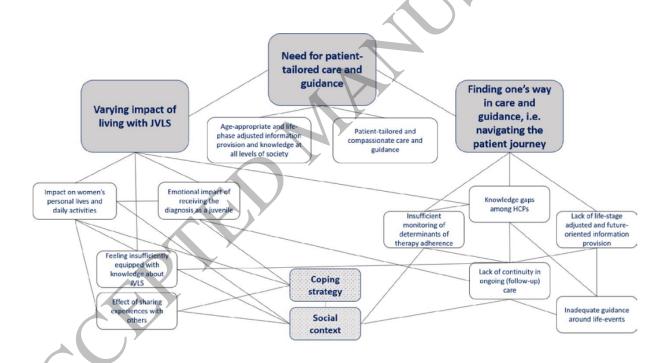


Figure 1. Thematic network of main- and subthemes and influencing factors regarding the experiences of women with a history of JVLS and their needs for care and guidance

The interrelationships between the (sub)themes are indicated by connecting lines.

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Figure 1 160x104 mm (DPI)

# Social context

# Varying impact of living with JVLS

Childhood

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2 3 Adolescence

Adulthood

Finding one's way in care and guidance

Need for patienttailored care and guidance

Coping strategy

Figure 2. Schematic overview themes: Experiences of women with a history of JVLS and their needs for care and guidance

The three phases of life (left, grey inner arrows), the two main themes (left, dark blue outer arrows), the two influencing factors (left, grey outer circle), resulting in the third, and overriding main theme (right, grey inner circle).

Figure 2 160x74 mm (DPI)

