



# Penile lichen sclerosus, circumcision and sequelae, what are the questions?

Beth Morrel<sup>1,2^</sup>, Lisette A. 't Hoen<sup>3^</sup>, Suzanne G. M. A. Pasmans<sup>2^</sup>

<sup>1</sup>Department of Obstetrics and Gynecology, Erasmus MC University Medical Center, Rotterdam-Sophia Children's Hospital, Rotterdam, The Netherlands; <sup>2</sup>Department of Dermatology, Center of Pediatric Dermatology, Erasmus MC University Medical Center, Rotterdam-Sophia Children's Hospital, Rotterdam, The Netherlands; <sup>3</sup>Department of Pediatric Urology, Erasmus MC University Medical Center, Rotterdam-Sophia Children's Hospital, Rotterdam, The Netherlands

*Correspondence to:* Beth Morrel. Department of Obstetrics and Gynecology, Erasmus MC University Medical Center, Rotterdam-Sophia Children's Hospital, Rotterdam, The Netherlands; Department of Dermatology, Center of Pediatric Dermatology, Erasmus MC University Medical Center, Rotterdam-Sophia Children's Hospital, Rotterdam, The Netherlands. Email: b.morrel@erasmusmc.nl

*Comment on:* Kwok M, Shugg N, Siriwardana A, *et al.* Prevalence and sequelae of penile lichen sclerosus in males presenting for circumcision in regional Australia: a multicentre retrospective cohort study. *Transl Androl Urol* 2022;11:780-5.

Submitted May 09, 2022. Accepted for publication Jun 15, 2022.

doi: 10.21037/tau-22-343

**View this article at:** <https://dx.doi.org/10.21037/tau-22-343>

Male circumcision as a religious or cultural rite has been performed for at least three thousand years, as seen in the ancient Egyptians' wall paintings and mummies (1). This tradition has been practiced in various communities on most continents throughout civilization, though different cultures perform ritual circumcision at different moments during the life-span, be it at birth as in Judaic tradition, 'bris', during childhood in some Muslim communities, 'khitan', or as a rite of passage into adulthood for the adolescent in some African tribal cultures, e.g., 'tahir' in Sudan (2,3). More recently, since the 19th century, medical therapeutic circumcision for conditions such as phimosis or recurrent infections have gained wide-spread use. In Africa, as well, circumcision is considered an important factor in reducing HIV-transmission (4). Thus, circumcision is one of the most common urological procedures performed on males of any age, be it motivated by ritual, religious or medical considerations (1).

One would expect our knowledge on phimosis which affects approximately 1% of all males (5) to be extensive. It is well established that phimosis is often due to penile lichen sclerosus (LS), formerly referred to as balanitis xerotica obliterans (6). Symptoms include non-retractile

foreskin, ballooning of the foreskin and problems with micturition. The prevalence of LS in males with phimosis undergoing circumcision is stated as anywhere between 2% to 95% in boys (7). However, histology is not routinely done, accounting for this large range. Remarkably, clinical suspicion of LS in boys seems to have a diagnostic accuracy of a mere 53% (6). Does this reflect a diversity in symptomology or the clinician being unaware of the possibility of LS? What is the true rate of LS when phimosis is present?

Some reports claim up to a 95% success rate of circumcision for phimosis, but this is surely an optimistic estimate, not accounting for long-term relapses or complications (8). Meatus stenosis or strictures are sometimes referred to as complication of circumcision, but with a rate of urethral strictures up to 20% seen in boys at the time of circumcision (9) it should be questioned if strictures are a consequence of the procedure or a complication of the LS itself. One could conjecture that a standard peri- or postoperative corticosteroid adjuvant therapy might help reduce the high incidence of post-operative strictures.

A declaration by the American Association of Pediatrics

<sup>^</sup> ORCID: Beth Morrel, 0000-0001-7609-725; Lisette A. 't Hoen, 0000-0002-4882-4311; Suzanne G. M. A. Pasmans, 0000-0003-1018-4475.

Taskforce on male circumcision published in 2012 (10) showed a shift toward a more positive attitude to neonatal circumcision, extolling the protection against sexually transmittable diseases and risk of penile cancer while stating no evidence of a negative effect on sexual well-being. However, a broad range of functions of the foreskin such as protection, erotic pleasure and promoting lubrication are given as arguments against routine circumcision, though the evidence has been described as being of 'low-level' (11) and less robust than studies on the advantages of circumcision. Considering these factors, could a nonsurgical approach to phimosis due to LS be tenable?

When treating women with vulvar LS, by comparison, excision of the lichenoid tissue is not an option. Nevertheless, this had been tried in the past. Why after removing the genital tissue affected with LS in females and replacing it with an autograft of healthy tissue from a distant site does the LS returns to haunt the subject at the graft site (12)? The local environment seems to be of paramount importance. After circumcision the local environment of the glans is changed which may be the reason that some men do not have a relapse. Genital LS in females is generally well-controlled with a regimen of potent steroids, first as treatment and then followed by maintenance application. No surgical procedure is necessary. And compliance to a maintenance regimen has even been shown to be protective against the development of vulvar squamous cell carcinoma (SCC) (13). Medical management has been proposed for penile LS, to conserve the prepuce, given the positive effect described for sensitivity, self-image and sexuality. However, the results and opinions vary (8,14,15).

Then there is the subject of genital SCC. Though in the general population a rare disease with an absolute risk of penile cancer of 0.0011% and an incidence rate of 1:100,000, for men with genital LS the absolute risk of developing a penile SCC is up to 0.91% (16), a more than 800-fold increase, with an incidence rate of up to 6.49/1,000 person years. For women the numbers are even higher with an absolute risk of developing a vulvar SCC in the presence of vulvar LS being up to 3.88% while the absolute risk in the general female population of a VSCC is 0.002% (16). Conversely, genital LS is found adjacent to vulvar SCC in more than half of the cases (17), and LS is found in 40% or more of the cases of penile SCC (18,19). The most recent European guidelines discuss the prevalence of non-HPV penile SCC in relation to LS (20). Circumcision in adulthood does not protect against penile cancer. In

contrast, among populations where early circumcision is routine, such as in Israel where circumcision ('bris') is performed on the 8th day after birth, the lowest prevalence of penile cancer is seen world-wide (20).

The article in the current issue of *Translational Andrology and Urology*, by authors Kwok *et al.* (21) from Australia, on a multi-centered retrospective study presents interesting population data regarding the histology of circumcision specimens and the sequelae to circumcision. Though their study does not give a definitive answer to the questions at hand, their research adds substantial data to information regarding prevalence of LS in males with phimosis. Histology was not standard in the participating clinics and the histology rates varied considerably between participating institutions. Could financial or cultural factors influence whether tissue specimens are analyzed? Since circumcision is so common, even if only for a limited amount of time such as a few years, our knowledge would be greatly serviced by advising standard histology of the tissue removed at circumcision. Combining this with population information about ritual or religious circumcision within the communities studied would yield robust data on the frequency of LS being found at circumcision for phimosis.

The questions now on the table that are imploring further studies are thus: what is the role of LS in causing phimosis; could a non-surgical treatment with potent corticosteroids be the first line of treatment with the goal of avoiding surgical intervention; and when surgery is performed should preoperative, postoperative and maintenance therapy with potent steroids be a part of standard treatment for penile LS? Finally, urologists treating penile LS may consider combining forces with gynecologists and dermatologists working in the field of LS to facilitate deeper insight into the etiology of genital LS, phimosis, the benefit and risks of circumcision, and the complications of genital LS as well as the occurrence of non-HPV related genital SCC. And visa versa, the work of gynecologists and dermatologist treating and studying genital LS would greatly benefit from collaboration with urologists for the same reasons.

Should we care if the questions posed above and by Kwok *et al.* (21) are answered? The basic tenant of medicine of non-maleficence, *primum non nocere*, may be kept in mind. Precisely because circumcision is such a pervasive procedure, its use should be founded on solid knowledge regarding the diseases for which it is performed, the efficacy of the procedure and the risks and benefits for those

undergoing this operation.

## Acknowledgments

*Funding:* None.

## Footnote

*Provenance and Peer Review:* This article was commissioned by the editorial office, *Translational Andrology and Urology*. The article did not undergo external peer review.

*Conflicts of Interest:* All authors have completed the ICMJE uniform disclosure form (available at <https://tau.amegroups.com/article/view/10.21037/tau-22-343/coif>). The authors have no conflicts of interest to declare.

*Ethical Statement:* The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

*Open Access Statement:* This is an Open Access article distributed in accordance with the Creative Commons Attribution-NonCommercial-NoDerivs 4.0 International License (CC BY-NC-ND 4.0), which permits the non-commercial replication and distribution of the article with the strict proviso that no changes or edits are made and the original work is properly cited (including links to both the formal publication through the relevant DOI and the license). See: <https://creativecommons.org/licenses/by-nc-nd/4.0/>.

## References

- Dunsmuir WD, Gordon EM. The history of circumcision. *BJU Int* 1999;83 Suppl 1:1-12.
- Irele FA, Jeyifo B, editors. *The Oxford Encyclopedia of African Thought*. Oxford: Oxford University Press, 2010.
- Morris BJ, Wamai RG, Henebeng EB, et al. Estimation of country-specific and global prevalence of male circumcision. *Popul Health Metr* 2016;14:4.
- Preventing HIV Through Safe Voluntary Medical Male Circumcision for Adolescent Boys and Men in Generalized HIV Epidemics: Recommendations and Key Considerations. Geneva: World Health Organization, 2020.
- Nguyen ATM, Holland AJA. Balanitis xerotica obliterans: an update for clinicians. *Eur J Pediatr* 2020;179:9-16.
- Celis S, Reed F, Murphy F, et al. Balanitis xerotica obliterans in children and adolescents: a literature review and clinical series. *J Pediatr Urol* 2014;10:34-9.
- Li J, Deng C, Peng Q. Underestimation of genital lichen sclerosis incidence in boys with phimosis: results from a systematic review. *Pediatr Surg Int* 2018;34:1245-50.
- Belsante MJ, Selph JP, Peterson AC. The contemporary management of urethral strictures in men resulting from lichen sclerosis. *Transl Androl Urol* 2015;4:22-8.
- Holbrook C, Tsang T. Management of boys with abnormal appearance of meatus at circumcision for balanitis xerotica obliterans. *Ann R Coll Surg Engl* 2011;93:482-4.
- American Academy of Pediatrics Task Force on Circumcision. Male circumcision. *Pediatrics* 2012;130:e756-85.
- Morris BJ, Moreton S, Krieger JN. Critical evaluation of arguments opposing male circumcision: A systematic review. *J Evid Based Med* 2019;12:263-90.
- Abramov Y, Elchalal U, Abramov D, et al. Surgical treatment of vulvar lichen sclerosis: a review. *Obstet Gynecol Surv* 1996;51:193-9.
- Lee A, Bradford J, Fischer G. Long-term Management of Adult Vulvar Lichen Sclerosis: A Prospective Cohort Study of 507 Women. *JAMA Dermatol* 2015;151:1061-7.
- Bossio JA, Pukall CF, Steele SS. Examining Penile Sensitivity in Neonatally Circumcised and Intact Men Using Quantitative Sensory Testing. *J Urol* 2016;195:1848-53.
- Morris BJ, Krieger JN. Letter from Morris and Krieger Re: Examining Penile Sensitivity in Neonatally Circumcised and Intact Men Using Quantitative Sensory Testing: J. A. Bossio, C. F. Pukall and S. S. Steele *J Urol* 2016;195:1848-1853. *J Urol* 2016;196:1824-5.
- Spekreijse JJ, Streng BMM, Vermeulen RFM, et al. The risk of developing squamous cell carcinoma in patients with anogenital lichen sclerosis: A systematic review. *Gynecol Oncol* 2020;157:671-7.
- van de Nieuwenhof HP, van der Avoort IA, de Hullu JA. Review of squamous premalignant vulvar lesions. *Crit Rev Oncol Hematol* 2008;68:131-56.
- Philippou P, Shabbir M, Ralph DJ, et al. Genital lichen sclerosis/balanitis xerotica obliterans in men with penile carcinoma: a critical analysis. *BJU Int* 2013;111:970-6.
- Powell J, Robson A, Cranston D, et al. High incidence of lichen sclerosis in patients with squamous cell carcinoma of the penis. *Br J Dermatol* 2001;145:85-9.
- Hakenberg OW, Compérat E, Minhas S, et al. EAU guidelines on penile cancer. European Association of

- Urology 2022. ISBN: 978-94-92671-16-5.
21. Kwok M, Shugg N, Siriwardana A, et al. Prevalence and sequelae of penile lichen sclerosus in males presenting

for circumcision in regional Australia: a multicentre retrospective cohort study. *Transl Androl Urol* 2022;11:780-5.

**Cite this article as:** Morrel B, 't Hoen LA, Pasmans SGMA. Penile lichen sclerosus, circumcision and sequelae, what are the questions? *Transl Androl Urol* 2022;11(7):910-913. doi: 10.21037/tau-22-343