

Original Research

A survey of adult men who underwent circumcision in childhood for pathological phimosis

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Abstract

Purpose: Pathological phimosis in childhood typically results in circumcision. Long-term follow-up data for men circumcised in childhood are lacking. This study sought long-term data regarding satisfaction with circumcision and voiding symptoms from men who had childhood circumcision between 1989 and 2010. **Methods:** Following ethical approval, a postal survey was sent to males > 17 years who underwent circumcision at an age < 16 years for pathological phimosis. **Results:** The survey was sent to 177 men, 23 completed surveys were returned [19 histology proven Lichen Sclerosus [LS, BXO], 4 chronic balanitis]. Mean age at circumcision was 9.7 years [range 3-15], at survey 23.5 years [18-37]. Seven [all > 9 years] remembered the decision for circumcision. Four warranted urologist review as adults, three required surgery for voiding symptoms and one still performs structure therapy; all had LS. When asked which treatment option[s] they would have considered; ten of them chose circumcision, nine of them chose preputioplasty, eight of them chose topical creams [two of them chose all three options, one man chose both foreskin preserving options]. Eight agreed with the statement "Having a circumcision in childhood or adolescence had an impact on my adult life"; this was positive in two men but negative in five men-they wouldn't have chosen circumcision. **Conclusions:** The response rate to the survey was low, making it difficult to draw firm conclusions. However, even in this limited sample, not all men would choose childhood circumcision for pathological phimosis, data supporting the need for larger studies of alternative treatment options. In addition, some men circumcised for childhood LS had significant voiding difficulties in adulthood.

Keywords

Balanitis xerotica obliterans; Child; Circumcision; Lichen Sclerosus et atrophicus; Surgical decision making

1. Background

In most boys a non-retractile foreskin, or physiological phimosis, is a natural stage in the development of the foreskin [1, 2]. However, approximately, 0.6% of boys under 15 years of age develop chronic balanitis or Lichen Sclerosus et atrophicus [LS] which may result in pathological phimosis [3, 4]. LS, also known as Balanitis Xerotica Obliterans [BXO],

is a recognised cause of pathological phimosis in children [2, 3]. Topical steroid ointment can be an effective treatment of pathological phimosis, though whilst mild LS responds well, topical therapy is less effective in more advanced disease [5]. If topical therapy fails or is considered inappropriate, circumcision is the recommended surgical treatment [4, 6]. Foreskin preserving surgical options, such as preputioplasty combined with intralesional steroid, have been described as

Childhood Circumcision Survey – please return your completed questionnaire in the envelope provided. If you prefer to complete this online please use this link:

<https://www.surveymonkey.com/s/YFHLYL2>

1. Please enter your study number, which is on the top of the letter

2. Can you recall the reason why you had a circumcision. Tick all reasons that you can remember:

- Foreskin did not go back Foreskin infections Urine infections
 Scarring of the foreskin Balanitis Xerotica Obliterans (BXO) Pain
 Religious reasons Not sure

3. Can you remember if you were included in making the decision to have the operation?

- Yes No can you tell me anything more about your answer?

4. Have you ever had to visit your GP or a hospital doctor / urologist (doctor specialising in urinary tract problems) with any of the difficulties from the list since your circumcision? Please tick all that apply.

	Not had to see	Fine or weak stream	Taking a long time to pass urine	Spraying stream	Pain passing urine	Urine infection
Visit to GP						
Referral or visit to a hospital doctor						

5. If you have had to see your GP or hospital doctor, how many times have you seen them about your urinary problems?

	1-2 times	3-5 times	6-8 times	More than 8 times
Number of times				

6. Have you had an operation or had to dilate your urethra (stricture therapy) to open up your urethral opening (the hole your wee comes out from) since your operation?

	No	Yes - once	Yes - twice	Yes - three times or more
Operation				
Stricture therapy				

7. Have you had any treatment to improve urine flow symptoms that did not involve an operation?

- No Yes - please describe

8. "Having a circumcision in childhood or adolescence had an impact on my adult life". To what extent do you agree with this statement?

- Strongly agree Agree Neither agree nor disagree Disagree Strongly disagree

If you chose agree or strongly agree can you explain why/how?

9. Within the last few years new treatments have become available that mean that some boys could avoid having a circumcision; if these treatments had been available when you were younger, what option may you have chosen? Tick all which apply.

- Circumcision A cut in the foreskin that widens the opening Local treatment with ointment to the foreskin

If you would like to share any further information about your experience please use the space below

10. By sending you this questionnaire, have we raised any worries or concerns? If so how might you manage those concerns?

- No concerns Talk to my GP Talk to my partner Talk to family or friends
 Other – please specify Contact the Research Study Team for advice

You can call the team on xxxx xxx xxxx. There is an answer phone for you to leave a message for the Research Team. The Research Team will return your call and signpost you to the right service to help answer your concerns.

FIG. 1. The survey.

an alternative to circumcision for boys with LS [2,7]. Emerging data shows that between one in four to one in five boys go on to have a circumcision, i.e., 75-80% of boys retain their foreskin [8]. However, foreskin preserving surgery has not been subject to sufficient investigation in the form of trials to be accepted as a mainstream management option. As a result, few surgeons offer this treatment and consequently evidence of both efficacy and long-term outcomes are lacking.

There is virtually no long-term data regarding the impact of childhood circumcision undertaken for pathological phimosis for men when they are older. Equally, there is no published long term data regarding sequelae of childhood LS such as meatal or urethral pathology [9]. In contrast, it is known that some men who undergo non-therapeutic circumcision as infants wish they had not been circumcised [10]. Circumcision may be the only surgical option given for management of pathological phimosis in children. However, not all families nor children want circumcision and some of those actively seek alternative treatments. Since there are alternatives, all be they poorly evidenced, further information is required to ascertain whether further research into the alternatives is called for. Equally it is important to ascertain what impact a circumcision has had for men who underwent the procedure as children. Therefore, this study aimed to ascertain long-term outcomes, both physical and psychological, in a population of men who had a circumcision for pathological phimosis in childhood.

2. Methods

An anonymous postal survey was sent to men who had circumcision for pathological phimosis in childhood. Ethical approval was obtained from the NHS Research Ethics Committee North West [REC reference 15/NW/0380, IRAS project ID 145578]. Ethical approval for contact by telephone and mail was applied for though permission was granted for contact by mail only. Funding was obtained from the hospital research fund.

2.1 Survey development and pilot

A set of 10 questions was designed. In order to determine acceptability and sensitivity, a random sample of male staff and visitors within the hospital were invited to complete the survey and comment on appropriateness and phrasing of the questions. Participants in the pilot study were both circumcised and non-circumcised and they could indicate circumcision state in their response if they wished. Minor amendments were made based on the results of the pilot: the final survey is shown in Fig. 1.

2.2 Participants and sampling

Male patients who underwent circumcision for pathological phimosis at a single UK tertiary paediatric centre from 1989 to 2010 were identified via hospital databases. The database for years 1989-2002 was a hand written record of every boy who underwent circumcision under the care of the single-handed paediatric urologist. From 2003 to 2010,

the database was collated from the electronic patient record system, searching by the code for circumcision and thus boys under the care of any surgeon in the hospital were included. The archived medical records were incomplete so other than the date of surgery and the histological analysis of the foreskin, no other data [such as pre-circumcision topical therapy, discussion of alternatives and risks] were available. Only those who were 18 years and over at the time of survey distribution and in whom there was a histological diagnosis of LS or chronic balanitis were eligible for the study. The study population was primarily Caucasian. Data regarding religion were not captured since the indication for circumcision was pathological phimosis. The circumcision rate in England around the time of the study was between 2.1 and 2.6 per 100 boys per year [11].

A paper copy of the survey was sent to eligible participants with a covering letter and a return, stamped and addressed envelope. The address for each patient was ascertained from the "NHS portal." A further copy of the survey was posted to those who had not replied 6-12 weeks after the first invitation. No patient identifiable data were included in the survey in order to maximise confidentiality.

3. Results

The survey was posted to 177 men, 138 with LS and 39 with chronic balanitis. In total, 27 surveys were returned by post. Four of the forms were blank, leaving just 23 for analysis.

The median age at circumcision of respondents was 9.7 years [range 3-15]. The median age at survey completion was 23.5 years [range 18-37] and the median number of years lapsed between circumcision and survey was 15 [range 5-24]. Nineteen men had LS and four had chronic balanitis.

When asked to recall their inclusion in the surgical decision-making process 9 men answered "yes"; the median age of this group was 13 years [range 10-15]. The median age of those answering "no" was 8 years [range 3-12]. Most men [19] recalled at least one symptom that had resulted in circumcision; the 4 that did not were all aged < 11 years.

Four men [17%] have had further surgery as an adult, three for voiding symptoms and one underwent a "redo" procedure. All had LS. One respondent, aged 12 years at the time of circumcision, has required three surgical procedures and continues to practice self dilatation [stricture therapy].

The survey provided respondents with three treatment options for pathological phimosis; they could consider choosing any or all of the treatments. When asked which options they may have considered in childhood, ten chose circumcision, nine chose preputioplasty and eight chose topical creams. Two men chose all three options, one man chose both foreskin preserving options and the remainder indicated just one option. There was notable difference in the age at circumcision between those choosing circumcision [median 8 years, range 3-14] and those choosing foreskin preserving options [median 12, range 10-14].

When considering the impact of childhood circumcision in adult life, eight respondents agreed or strongly agreed with the statement "Having a circumcision in childhood or ado-

lescence had an impact on my adult life.” Two respondents commented that it had positively impacted their adult life, and would choose circumcision again whilst five made comments implying a negative impact and each of them choose foreskin preserving options in the preceding question. Eight men indicated that they neither agreed nor disagreed with the statement and three disagreed although none of these men made comments so their reasons for choosing this option are unclear.

Further, 12 respondents used the free text spaces. Comments from those who seemed positive about their childhood circumcision included; “decision was made for my benefit,” “more healthy,” “circumcision is cleaner, don’t know differently if done younger,” “solved problems before they could affect adult life” and “worried as an adolescent, but not now.”

For those respondents who felt negatively, the comments were generally longer and in greater detail. For example: “I think about it regularly, have a low self-esteem, embarrassed, don’t make decision for children”; “I didn’t have a choice, it’s painful, impacts on my self-esteem”; “there is a loss of sensitivity and I feel it has affected my sexual function”; “scars that require hygiene”; “my sex life isn’t as good and would avoid operation if possible”; “I’m trying to regrow my foreskin, stop circumcising children” and “the doctor made me feel like I had no choice, biggest regret of my life.”

4. Discussion

Circumcision is currently the mainstay of treatment for pathological phimosis in childhood; indeed, it is considered to be the “gold standard” treatment for LS [4, 6]. However, there are no longitudinal outcome studies of males who have undergone therapeutic/medical childhood circumcision, nor of men’s attitudes towards “medical” circumcision undertaken when they were children [9]. Our intention was to use the results from this survey to inform future studies of surgical treatment options for pathological phimosis in children. Unfortunately, the survey response rate was very low, such that firm conclusions are hard to draw. The nature of the ethical approval for this study did not give scope to extend the study in any way, nor to explore the reasons behind the poor response rate as it was considered inappropriate to contact eligible men in any other way. The survey was deliberately short as it was felt that participants would be more likely to complete it if it were short [12]. That meant that we were unable to explore other factors which may influence men’s perceptions of their circumcision such as their religion, sexuality, partnership status and factors such as the views of their social circle or community around the acceptability of circumcision.

Nevertheless, we believe the responses received are still worthy of scrutiny as no-one has ever published the results of an attempt to seek such data before. The low response rate in itself suggests that this is a topic many men prefer not to consider. Perhaps, this is because their circumcision was done a long time ago and they do not think about it and the survey may have seemed pointless—such men probably have neutral or positive perceptions of their circumcision. On

the other hand, it is likely that some men did not reply as they did not wish to dwell on negative perceptions they have regarding their circumcision.

The findings of the study, whilst limited, revealed a range of attitudes: some men were satisfied with their childhood circumcision, some were neutral about the procedure and there were a number of men who had clear negative views of their circumcision. There was also a small cohort of four men who had experienced voiding symptoms requiring surgical intervention as adults. It is impossible to know whether their underlying diagnosis of LS or the circumcision was the causative factor and both could be conceivable. It is well documented that boys with LS may develop metal stenosis and older men with LS can experience urinary [and sexual function] symptoms [13, 14]. Thus, the reports from men with childhood LS requiring intervention as adults is not that surprising. It is notable that there are no publications recording late complications of circumcision for pathological phimosis in childhood that we could identify.

Several of the respondents disclosed that they felt that their circumcision had affected their sex life and self-esteem, and similar disclosures are made by some men who have undergone neonatal non-therapeutic circumcision [10]. However, this is a controversial subject and there are many publications both supporting and refuting the potential impact of circumcision. A recent study by Bronselaer *et al.* argued that loss of the foreskin decreases glans sensation and sexual pleasure but their methodology was criticised and a recent systematic review concludes that circumcision has no adverse effect on penile sensation based on histology and correlates of sexual function [15–17]. Much of the literature on the subject focuses on non-therapeutic circumcision in neonates [18]. It is unknown if the combination of foreskin pathology and circumcision in childhood might influence feelings of sexual inadequacy in adulthood in a similar way to that reported by men who underwent non-therapeutic circumcision in their early years [19, 20].

The majority of boys with LS are treated by circumcision and in this study, we identified a smaller cohort undergoing circumcision for chronic balanitis [8]. The long-term perceptions expressed by the respondents to this survey show that surgeons should be mindful about how to best counsel boys and their parents. Common sense would suggest that younger boys are less likely to recall the decision for surgery and the responses seem to support this. Interestingly, of the eight men who did recall being involved in the decision for surgery, only one indicated that he would chose circumcision over foreskin preserving options if offered the choice now. The remainder would have chosen foreskin preserving options over circumcision. From these responses one might consider that circumcision at a younger age, when boys do not remember the decision and the reasons for it, seems to be associated with less regret than decisions for circumcision in older boys when they were more aware of the discussion surrounding surgery. Indeed, a larger Belgian study found a difference between those circumcised before or after the age of 10 years, with boys circumcised during adolescence

reporting more negative associations with circumcision than those circumcised at a younger age [15].

Many children have the capacity and the capability to be included in decision-making about their health and age may be an important consideration when exploring inclusion of the boys themselves in surgical decision-making [21]. Shared decision-making is essential to patient centred care [22]. Therefore, we believe that surgeons should explore non-surgical as well as foreskin preserving options, especially when counselling older boys.

5. Limitations

The sample size was good however the response rate was very low and it is well known that postal questionnaires yield a poor return rate [23]. There are a number of likely reasons for this, the simplest of which is that not all men received the survey. A small number of surveys were returned marked "unknown at this address" and others may have been discarded if the name was not recognised by the current occupant of the property. Whilst the accuracy of the "NHS portal" should be high, it is not guaranteed as a person's address is only updated when the General Practitioner is informed. If a patient moves house and they are in good health, they may well postpone registration with a new GP.

The nature of the questions, the unexpected contact and the time from surgery may have also been the factors that limited participation as alluded to above. Finally, as we were asking some participants to think back over two decades they may have felt unhappy to try and recall events from such a long time before. While the team originally planned telephone contact with eligible participants, the Research Ethics Committee considered that to be too intrusive and they advised that a postal questionnaire should be used as the only method of contact as this could be more readily ignored or dismissed. Due to the possibility of causing concern by contacting men unexpectedly, the survey included advice directing them to support if they had any concerns as a result of the request to participate.

6. Conclusions

Not all men would choose childhood circumcision as first line management for pathological phimosis suggesting that further studies of alternatives are required. Our study highlights the need to understand the longer-term outcome for men who experience pathological phimosis as children and the psychosexual consequence of LS and circumcision in later adult, male life should not be dismissed. Surgeons need to be mindful of the capacity of boys from as early as 8 years of age, encouraging active involvement in the surgical decision making process for treatment of pathological phimosis. Furthermore, men with childhood LS need to be aware of complications that may occur in adulthood, thereby promoting early health seeking behaviours.

Ethics approval and consent to participate

Ethical approval was obtained from the NHS Research Ethics Service Committee North West [REC reference 15/NW/0380, IRAS project ID 145578].

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Conflict of interest

None of the authors have any conflict of interest to declare.

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