

Foreskin Morbidity in Uncircumcised Males

Ida Sneppen, MD,^a Jørgen Thorup, MD, PhD^{a,b}

abstract

OBJECTIVE: As a consequence of the discussion on whether the health benefits of newborn male circumcision outweigh the risks and the discrepancies in reported figures of complications, we evaluated the incidence and morbidity of foreskin surgery due to medical indications in boys from the Capital Region of Denmark in 2014.

METHODS: Medical records from all boys operated on the foreskin due to medical reasons in the Capital Region in 2014 were reviewed. Patients with hypospadias, ritual circumcision, and redo-surgery because of complications to nontherapeutic circumcision were excluded.

RESULTS: A total of 181 patients were included. The cumulative risk of undergoing foreskin operation before 18 years of age was 1.7%. Forty patients had histologic verified balanitis xerotica obliterans (BXO) corresponding to a total risk of 0.37% of developing BXO. Mean age at surgery was 10.1 years (range 1–17). Phimosis was the most frequently reported indication (95.0%). The remaining 5.0% underwent surgery because of frenulum breve causing problems during erection. Before surgery, 27.1% had foreskin-related voiding problems and 17.1% had at least 1 episode of balanitis. Circumcision was initially performed in 44 cases. The remaining 137 patients had a foreskin-preserving operation performed. Nine boys had secondary circumcision after initially having foreskin-preserving operation. Fifty patients initially had preputial histology performed. BXO was verified in 37 patients. Of the 9 patients with redo-surgery due to recurrent phimosis, a further 3 had histologically verified BXO.

CONCLUSIONS: Childhood foreskin-related problems in a region with no tradition of newborn male circumcision should not be neglected.

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Dr Sneppen carried out the initial analyses and drafted the initial manuscript; Dr Thorup conceptualized and designed the study, and reviewed and revised the manuscript; and both authors approved the final manuscript as submitted and agree to be accountable for all aspects of the work.

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WHAT'S KNOWN ON THIS SUBJECT: The risk of undergoing foreskin surgery due to medical reasons is in accordance with the few studies of the literature varying from 0.6% to 2.1%.

WHAT THIS STUDY ADDS: A surprisingly high incidence and morbidity related to phimosis in boys from a well-defined catchment area where a strict foreskin-preserving culture is dominating could be demonstrated. This is relevant for the debate on newborn male circumcision.

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The practice of circumcising boys is carried out for 3 major reasons: (1) due to acute medical indications, (2) as a cultural (also known as nontherapeutic) practice, and (3) to prevent later diseases possibly affecting the foreskin.^{1,2} During the later years, the latter 2 reasons have been under much debate. A recent report published in *Pediatrics* from the American Academy of Pediatrics concludes the following: "Evaluation of current evidence indicates that the health benefits of newborn male circumcision outweigh the risk, and the benefits of newborn male circumcision justify access to this procedure for those who choose it."² In contrast, a large group of European pediatric urologists, pediatric surgeons, and pediatricians argued in a letter to the editor that the aforementioned article is strongly culturally biased and that circumcision fails to meet the commonly accepted criteria for the preventive medical procedures in children.³ In Denmark and several other countries in Europe, it has been considered to prohibit circumcision for nonmedical indication because it is regarded as a violation against the child according to the United Nations' Declaration of the Rights of the Children.^{1,4}

In the debate on circumcision, the reduced risk of urinary tract infection in the first years of life is the benefit argument for circumcision most often used for the pediatric age group.² However, few studies have investigated the morbidity related to the incidence of phimosis and results of foreskin surgery in childhood. In a recent Danish cross-sectional study of military recruits from 1996 to 2010, the self-reported incidence of treatment of phimosis was 3.9%. The type of treatment was not specified.⁵ A Brazilian study found that 2.1% of all Brazilian boys aged 1 to 14 years treated in the Brazilian unified health system had to undergo circumcision for medical reasons.⁶ The rate of

therapeutic circumcision in literature seems to be increasing.⁷

Our study differs from other studies due to the strict foreskin-preserving culture in Denmark. In general, circumcision is performed in Denmark only when medical treatment and other surgical techniques are considered inadequate.

The aim of this study was to evaluate the morbidity related to medically indicated foreskin operations in boys aged 0 to 17 years from a well-defined catchment area.

METHODS

In 2014, all boys in the Capital Region of Denmark aged 0 to 17 years with foreskin-related problems and possible need of surgery were referred to the Department of Pediatric Surgery, Rigshospitalet, University of Copenhagen. From the regional database, the surgical codes according to the Nordic Classification System of Surgical Procedures related to foreskin operations were used to identify the patients. We included KKGH00 sutura penis, KKGH10 operatio pro phimosis, KKGH80 operatio plastica cutis penis, KKGV00 repositio paraphimosis, KKGV10 desaglutinatio preputii, and KKGV20 circumcisio performed from January 1, 2014, to December 31, 2014. Due to local restrictions, unspecified codes were not available for documenting surgery. All Danish citizens have a unique social security number, which is linked to health care-related contacts. Using the social security number, we collected the matching medical records on all hospital contacts from the Danish hospital file system.

Demographic data of the boys were registered and patients referred from outside the Capital Region of Denmark were excluded to estimate the true risk for boys having

TABLE 1 Birth Rates of Living Boys in the Capital Region of Denmark in the Period 1996 to 2014

Year	Birth Rate
1996	11 118
1997	11 127
1998	10 949
1999	10 957
2000	11 104
2001	10 927
2002	10 241
2003	10 981
2004	10 983
2005	10 927
2006	11 047
2007	10 934
2008	10 962
2009	10 870
2010	11 247
2011	10 413
2012	10 604
2013	10 281
2014	10 639

operation on the foreskin in the birth cohort due to medical reasons.

Patients with concomitant diagnosis of hypospadias were excluded from the cohort. Furthermore, boys in need of additional surgery due to complications after nontherapeutic (cultural) circumcision were excluded. Nontherapeutic circumcision is not performed in the public health care system of Denmark.

Boys operated before 2014 and with redo-surgery in 2014 were also excluded.

To investigate the percentage of the birth cohort needing foreskin operation due to medical reason, we collected birth statistics. The size of the Danish male birth cohort 1996 to 2014 in the Capital Region of Denmark was found by using Danish Statistics (Table 1).⁸

To investigate the incidence of foreskin operations due to medical indications we divided our patient population by birth year from 1996 to 2014. Data from the full annual birth cohort from 1996 to 2014 were used to calculate incidence rate. The yearly male birth rate varied in the period between 10 241 and 11 247

(Table 1). Confidence intervals (CIs) were calculated by using McCallum Layton Stats Calculator (<https://www.mccallum-layton.co.uk/tools/statistic-calculators/confidence-interval-for-proportions-calculator/>) and checked with Geigy Scientific Tables. The study was conducted according to the Helsinki II declaration.

RESULTS

In 2014, a total of 581 boys were admitted to the Department of Pediatric Surgery because of phimosis-related problems. This figure lies between 5.2% (581/10 241) and 5.7% (581/11 247) of the annual birth cohort (1996–2014) (Table 1). Operation of the foreskin was performed on 207 (35.5%) of the referred patients.

After reviewing the patient charts, 26 patients were excluded. Two boys underwent ritual circumcision during operations for non-foreskin-related reasons (inguinal hernia and biliary atresia) for the boy to benefit from full anesthesia when the parents requested circumcision anyway. Five had redo-surgery for complications due to a nontherapeutic circumcision performed elsewhere. Three patients had hypospadias as a concomitant disease. One patient was not from the Capital Region of Denmark. Fifteen of the patients were admitted to redo-surgery after an operation done before 2014 and now in need of additional surgery. Therefore, 181 boys were included in our study.

Among the 181 boys included in our study, 100 (55.2%) patients had tried topical steroid application treatment in advance. Topical steroid application is often used as a “first choice” treatment of phimosis by general practitioners according to Danish guidelines. It was not possible to evaluate the treatment compliance of the patients and parents in this respect.

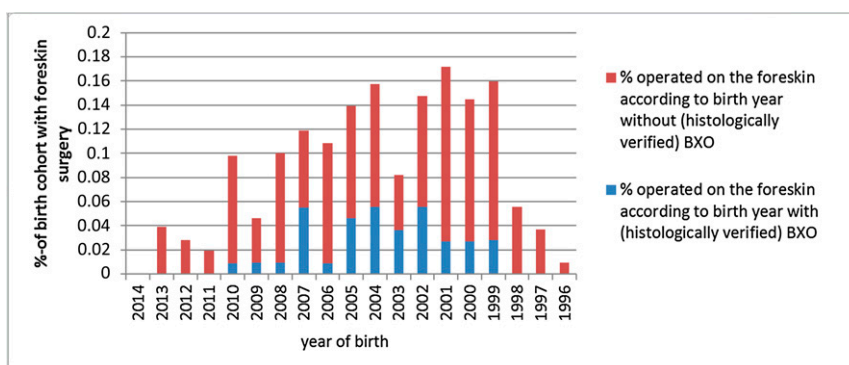


FIGURE 1

Percentage distribution of the 181 boys operated on the foreskin as a function of the cohort size of the corresponding year of birth. The size of the birth cohort varied between 10 241 and 11 247 births per year from 1996 to 2014 (Table 1).

Several patients had multiple problems related to their foreskin. Extracting all information from patient files, we found that 172 (95.0%) suffered from phimosis. Nine boys (5.0%) suffered from frenulum breve mainly giving symptoms during erection, and 49 (27.1%) patients had severe voiding problems before their operation, such as an insufficient flow or pain when urinating. Ballooning of the foreskin was not considered as a voiding problem. Forty-six patients (25.4%) were suspected of having balanitis xerotica obliterans (BXO) before surgery. Thirty-one boys (17.1%) had at least 1 episode with balanitis. Two (1.1%) patients had urinary tract infection. Three boys (1.7%) had suffered from paraphimosis.

The mean age at the time of surgery was 10.1 years (1 year–17 years), with a median of 11 years old. Follow-up data were obtained for all patients ranging from 7 to 20 months.

The age distribution among the patients compared with the size of the birth cohort is seen in Fig 1. We found the cumulative risk of undergoing a foreskin surgical procedure in general anesthesia before 18 years of age to be 1.66%. Furthermore, the percentage of boys with histologically verified BXO in our birth cohort was 0.37%. The age distribution of patients with

histologically verified BXO was 4 to 16 years.

Figure 2 illustrates the number and surgical procedure performed. All procedures were performed as day care surgery in general anesthetics with sacral block. Paracetamol was used for postoperative pain treatment. There were no anesthetic complications.

Forty-five patients (24.8%; 95% CI 18.5–31.1) contacted the surgical department due to complications after the operation with recurrent phimosis being predominant. Twelve (6.6%; 95% CI 3.0–10.2) of the inquiries were handled on the telephone and the patients had no further need to see a doctor. Thirty-three (18.2%; 95% CI 12.6–23.8) of the patients were readmitted to the in-ward hospital for treatment. The type of complication and eventual treatment are outlined in Table 2. Fewer than half of the 33 patients had >1 complication (data not shown).

In total, 10 patients (5.5%; 95% CI 2.2–8.8) had redo-surgery on the foreskin. Among the 44 patients circumcised, 1 needed redo-surgery on the foreskin due to an insufficient circumcision. The remaining patients who underwent redo-surgery suffered from a combination of recurrent phimosis and inflammatory changes. In the group of not primarily

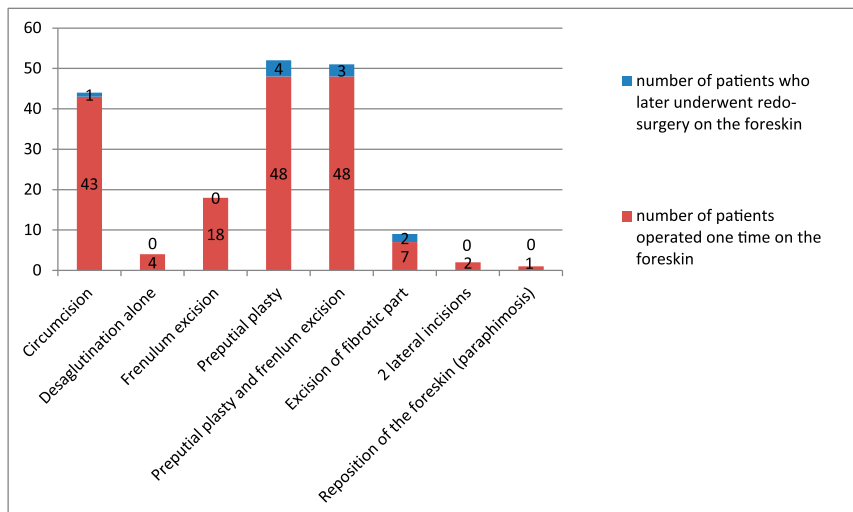


FIGURE 2

Type of operation primarily performed. Eighteen (9.9%) of the patients suffered only from frenulum breve and excision of the frenulum was the only action; 51 (28.2%) of the patients suffered from both frenulum breve and phimosis, and were treated with frenulum excision and preputial plasty. Phimosis alone indicated preputial plasty in 52 (28.7%). If the doctor suspected secondary phimosis because of BXO, circumcision would be the clinical guideline, which was the case for 44 (24.3%) patients. A smaller fraction consisting of 9 patients (5.0%) had a fibrotic part of the preputium excised instead of circumcision.

TABLE 2 Causes of Complications and Eventual Treatment After the Primary Surgery

	Inquiries Handled by Telephone	Readmitted to Hospital	In Total
Re-phimosis, <i>n</i> = 17			
No further treatment	2	4	6
Topical steroid only	0	2	2
Redo-surgery	0	10	10
Pain, <i>n</i> = 10	3	7 (+3 after redo-surgery)	10 (+3 after redo-surgery)
Bleeding, <i>n</i> = 12			
Compression	5	5	10
Topical adrenalin gauze compression	0	2	2
Swelling of the penis, <i>n</i> = 8	3	5	8
Voiding difficulty <10 d postoperative, <i>n</i> = 10			
Pain relieving/No treatment	0	6	6
Catheterization	0	4	4
Bad cosmetic result, <i>n</i> = 2			
No redo-surgery	0	2	2
Redo-surgery	0	0	0
Infection, <i>n</i> = 2			
Local antibiotics	0	1	1
Systemic antibiotics	0	1 (+1 at redo-surgery)	1 (+1 at redo-surgery)
Meatus stenosis, <i>n</i> = 19			
Only flow measures	0	6	6 (5.0%)
Meatus dilation (self)	0	1	1 (0.5%)
Meatus dilation (in general anesthesia)	0	12	12 (6.6%)

circumcised patients, 9 patients (6.6%) were later circumcised due to the aforementioned complications. Redo-surgery was required in 2 of 9 patients operated with excision of the fibrotic part of the preputium only. Both of the patients had recurrent phimosis due to BXO in the remaining foreskin. One of the patients primarily operated with frenulum excision and preputial plasty and later circumcised was suspected to have BXO, but circumcision was not done primarily because the parents had chosen a foreskin-preserving operation. Of the patients not suspected to have BXO at the first operation, 6 (4.4%) needed redo-surgery.

In our study sample, 49 (26.6%; 95% CI 20.2–33.0) boys were preoperatively suspected to have BXO. Of the 49 patients, 44 were examined with histology. In 36 patients, BXO was verified by histology, 3 patients had an inconclusive histology, and in 5 patients, no BXO, but chronic inflammation was found. In the remaining 5 cases, no histology was performed. Two of the cases with suspected BXO, but without histology, later developed meatus stenosis and required meatus dilation in general anesthesia, strongly suggesting an underlying BXO.

In 7 cases, the surgeon had not specified whether the preputium looked suspicious of BXO. In this group, 4 were histologically examined; 1 had BXO, 1 did not have BXO, and 2 were inconclusive, but 1 of those later developed BXO and required redo-surgery.

Among the 125 patients not suspected to have BXO, only 2 were examined histologically; 1 did not have BXO and the other was inconclusive. Another 2 patients later underwent redo-surgery and BXO was confirmed.

In total, 40 patients were histologically verified to have BXO. This corresponds to a risk of

TABLE 3 Concomitant Diagnosis

Type of Disease	<i>n</i>
Lung disease	
Interstitial pulmonary disease	1
Congenital cardiac malformations	3
Genito-urinary tract disease	
Cryptorchidism	10
Congenital malformation of the urinary tract	5
Gastrointestinal diseases	
Crohn disease	1
Anal prolapse	1
Biliary atresia	1
Reproductive organs	
Hydrocele	4
Penile hemangioma	1
Cyst on glans	1
Dermatologic diseases	
Cutis marmorata telangiectasia congenita	1
Psoriasis	3
Other malformations	
Cleft-lip-gum and palate	2
Deformity of outer ear	1
Vision/eye problems	4
Psychiatric	
Tourette	1
ADHD and ADD	7
Anxiety disorder	1
Autism (5 with infantile autism)	9
ASD not specified as autism (F84.8, 84.5)	3
Delayed language and psychomotor development	1
Cognitive deficits and developmental delay	1
Other	
Klinefelter syndrome	1
Di George syndrome (22q11 deletion syndrome): heart malformation, immunodeficiency, autism, and mental retardation	1
Myelomeningocele, hydrocele, detrusor hyperreflexia, hofstedysplasi	1
Diabetes type I	1
Cerebral palsy	1
Atopy (asthma/atopic dermatitis/rhino conjunctivitis)	7

ADD, attention-deficit disorder; ADHD, attention-deficit/hyperactivity disorder.

0.37% of developing BXO (Fig 1). Furthermore, 2 patients suspected to have BXO who were not histologically examined later developed meatus stenosis, strongly indicating an underlying BXO.

In total, 19 patients (10.5%; 95% CI 6.0–15.0) had meatus stenosis verified by abnormal flow measures. This corresponds to a risk of 0.17% of developing meatus stenosis before 18 years of age. Thirteen patients were treated with meatus dilation: 12 in general anesthesia and 1 patient was trained in self-dilation. The remaining 6 patients were followed with flow measures, but the stenosis did not require surgery. All 19 patients were suspected to have

BXO, but it was histologically verified in only 15 of the patients. In 3 cases, histology was not made and in the fourth case, histology description was inconclusive due to insufficient size of material. Comparing the 15 patients with meatus stenosis and histologically verified BXO with the 40 patients with histologically verified BXO in our study reveals that 37.5% of patients with histologically verified BXO developed meatus stenosis and 25.0% needed treatment of the stenosis.

Concomitant diseases were registered in 67 (37.0%; 95% CI 30.0–44.0) patients. Four patients had 2 concomitant diseases and 2 patients had 3 concomitant

diseases (data not shown). All of the concomitant diseases and the number of patients with each disease are shown in Table 3.

DISCUSSION

In the Capital Region of Denmark in 2014, we calculated a risk of 1.66% of undergoing a foreskin surgical procedure in general anesthesia before 18 years of age.

Due to the higher prevalence of nontherapeutic circumcision in many other countries, it would be difficult to estimate the real need of operation on the foreskin due to medical reasons in those countries. We did not subtract the number of boys who had undergone a nontherapeutic circumcision from the birth cohort. Therefore, the calculated risk of 1.66% for boys to develop pathologic phimosis in need of surgery might be slightly underestimated because a smaller fraction of the Danish population is Jewish and Muslim. This number is unknown, because the religious affiliation of a child is not registered. Private nontherapeutic circumcision is legal, but not registered, so the number of boys undergoing nontherapeutic circumcision is unknown in the Capital Region of Denmark. The Danish Health and Medicines Authority estimated a cultural (nontherapeutic) circumcision number of 1000 to 2000 boys per year in all of Denmark (based on the number of mothers with origin from a country where cultural circumcision is dominating).⁹ If the distribution of immigrants from such countries in Denmark were even, this would make a number of 310 to 620 undergoing a nontherapeutic circumcision per year in the Capital Region of Denmark. This is probably a slight underestimation because a higher fraction of Muslims and Jews settled in the capital city. We were not able to determine the number of people moving to and from the

Capital Region of Denmark in the background population, but a fair estimation would be an equal amount of relocations to and from.

Our calculated risk of 1.66% undergoing foreskin surgery due to medical reasons is in accordance with the few other studies estimating this percentage varying from 0.6% to 2.1%.^{6,10} Nonetheless, these figures should be taken in to account before nontherapeutic circumcision is prohibited.

At least 100 boys (55.2%; 95% CI 48.0–62.4) of the 181 patients included had used topical steroid treatment before the operation. This number might be larger due to incomplete preoperative notes from the pediatric surgeon making the examination. Correct use of a topical steroid is shown to reduce the need for surgery in 70% to 80% among patients with unscarred phimosis.¹¹ The success rate differs when the foreskin is affected by BXO: ranging from significant improvement in 18% after 3 months in foreskin mildly affected by BXO to no significant improvement in patients with established scar formation of the foreskin.¹²

Despite the unknown patient compliance with topical steroid therapy, which was not monitored systematically, we must assume that a larger number of boys responded well to steroid treatment and thus were not included in the 581 boys referred to the pediatric surgical department, but still had previous foreskin-related problems. So, also, the total number of patients in the Capital Region with foreskin-related problems may be underestimated.

The operation performed depends on the indication of the operation. In general, circumcision was chosen if the surgeon suspected BXO. When BXO is not suspected, one can choose to do either circumcision or foreskin-preserving surgery with a risk of needing redo-surgery. Nine (6.6%) of our patients who underwent a

foreskin-preserving operation were later circumcised due to recurrent phimosis.

Excision of only the fibrotic part of the foreskin was followed by need for redo-surgery in 2 (22.2%) of 9 patients, which supports the current clinical guidelines that a circumcision should be performed when BXO is suspected.^{13,14}

It is in accordance with the literature that 22.1% of our surgical material had BXO (verified by histology). In a review from Celis et al,¹⁵ the calculated incidence of BXO was 35% (variation 5.0% to 52.6%) among circumcised children.

Among all patients operated on the foreskin, 19 (10.5%; 95% CI 6.0–15.0) were verified to have meatal stenosis. All 19 patients were suspected to have BXO, but it was histologically verified in only 15 of the patients. Fifteen (37.5%) of 40 patients with histologically verified BXO developed meatus stenosis and 10 (25%) patients needed treatment of the stenosis. The true percentage of boys with BXO who develop meatus stenosis is most likely even higher in our study due to insufficient histologic examination. The percentage of boys requiring treatment of meatus stenosis among boys with histologically verified BXO is comparable to Homer et al,¹⁶ who found that 20% of boys with BXO require operation for meatal pathology during circumcision and a further 20% require a later meatal procedure.

We calculated the risk of developing meatus stenosis before 18 years of age in an uncircumcised population to be 0.17%, which is ~3 times lower than the incidence described in boys after newborn male circumcision.^{17,18} Few and minor anesthetic complications were described in our previous series on cultural circumcision, and in our present study there were none.¹⁷ Few boys were readmitted to in-ward hospital

treatment because of insufficient pain treatment (Table 2). We had no information about the time away from work for the parents and the time away from school or other activities for the patients.

In our patient population, we found that 7.2% (95% CI 3.4–11.0) were diagnosed with autism spectrum disorder (ASD) as a concomitant disease (Table 3). A recent Danish study calculated the prevalence of ASD in Denmark to be 1.5% among boys,¹⁹ but a higher prevalence of ASD in other background population studies has recently been calculated to be in accordance with our findings.²⁰ In a national cohort study, Frisch and Simonsen¹⁹ found an increased risk of autism in children having undergone ritual circumcision, hypothesizing that the procedure could be a precipitating factor in the development of autism. There could be no such confounding in our study, as the diagnosis of autism was known before the surgical procedure.

The prevalence of the other disorders is probably in accordance with the prevalence in society in general.

CONCLUSIONS

Our study shows significant morbidity related to foreskin problems in a predominantly uncircumcised population. More than 5% of boys in the catchment area were admitted to the pediatric surgical department with foreskin-related problems and at least 1.66% of the boys needed surgical procedures in general anesthesia with a 5.5% redo-surgery rate. These figures have to be taken under consideration when counseling parents about newborn male circumcision.

ABBREVIATIONS

ASD: autism spectrum disorder
BXO: balanitis xerotica obliterans
CI: confidence interval

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