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PRACA ORYGINALNA
ORIGINAL PAPER

The incidence of balanitis xerotica obliterans in patients surgically treated for phimosis

Częstość występowania *balanitis xerotica obliterans* u pacjentów leczonych chirurgicznie z powodu stulejki

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ABSTRACT

INTRODUCTION: Phimosis is a disease entity that society has given negative connotations. Discussions about the foreskin, both in the context of its pathological and physiological conditions have been going on for centuries, and include issues related to religion, hygiene, esthetics and tradition. In 2002, researchers from London recognised that secondary phimosis corresponds to lichen sclerosus et atrophicus, better known as balanitis xerotica obliterans (BXO), constituting it the only absolute indication for circumcision in boys. This study aims to assess the histopathological examinations of patients after the surgical treatment of phimosis in order to ascertain the relationship between BXO and clinically diagnosed phimosis.

MATERIAL AND METHODS: This study analyzed the results of histopathological examinations in patients who underwent surgical methods of phimosis treatment in the period from January 2014 to March 2020. The inclusion criteria of the study were the surgical treatment of phimosis with the accompanying histopathological examination of the specimens collected during the surgery. The data were collected prospectively and randomly.

RESULTS: There were 106 patients in whom a surgical procedure and histopathological examination were performed. The mean age of the patients was 9.41 ± 3.82 years. In the entire group of patients, the diagnosis of BXO (including BXO focal) was 59% (n = 63).

CONCLUSIONS: The most common cause of secondary phimosis after surgical treatment is BXO, with a patient prevalence ranging from 5.5% to 84%. The results of this study fall within this range. Visible scarring of the foreskin may also be caused by other factors such as chronic inflammation of the foreskin, but also poor hygiene or infections in this area.

KEYWORDS

BXO, balanitis xerotica obliterans, phimosis, lichen, impetigo, circumcision, balanitis, lichen sclerosus planus

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STRESZCZENIE

WPROWADZENIE: Stulejka jest jednostką chorobową, której społeczeństwo nadało negatywne konotacje. Dyskusje dotyczące napletka, zarówno w kontekście jego stanów patologicznych, jak i fizjologicznych, toczą się od wieków i obejmują zagadnienia związane z religią, higieną, estetyką oraz tradycją. W 2002 r. londyńscy badacze zidentyfikowali stulejkę wtórną jako odpowiadającą liszajowi twardzinowemu i zanikowemu (*lichen sclerosus et atrophicus*), znanemu również jako *balanitis xerotica obliterans* (BXO), który jest jedynym bezwzględnym wskazaniem do obrzezania u chłopców. Niniejsze badanie ma na celu ocenę badań histopatologicznych pacjentów po chirurgicznym leczeniu stulejki, aby ustalić związek między BXO i klinicznie rozpoznaną stulejką.

MATERIAŁ I METODY: W niniejszym badaniu przeanalizowano wyniki badań histopatologicznych u pacjentów poddanych chirurgicznym metodom leczenia stulejki w okresie od stycznia 2014 r. do marca 2020 r. Kryterium włączenia do badania było chirurgiczne leczenie stulejki z towarzyszącym badaniem histopatologicznym próbek pobranych podczas operacji. Dane zostały zebrane prospektywnie i losowo.

WYNIKI: W badaniu wzięło udział 106 pacjentów, u których przeprowadzono zabieg chirurgiczny, a uzyskane próbki poddano badaniu histopatologicznemu. Średni wiek pacjentów wynosił 9,41 ± 3,82 roku. W całej grupie pacjentów rozpoznanie BXO (w tym BXO *focalis*) wynosiło 59% (n = 63).

WNIOSKI: Najczęstszą przyczyną stulejki wtórnej po leczeniu operacyjnym jest BXO, którego częstość występowania u pacjentów wynosi od 5,5% do 84%. Wyniki niniejszego badania mieszczą się w tym zakresie. Widoczne bliznowacenie napletka może być również spowodowane innymi czynnikami, takimi jak przewlekły stan zapalny napletka, ale też niewłaściwą higieną czy zakażeniami tej okolicy.

SŁOWA KLUCZOWE

BXO, *balanitis xerotica obliterans*, stulejka, liszaj, napletek, obrzezanie, *balanitis*, liszaj twardzinowy płaski

INTRODUCTION

Phimosis is a disease entity that society has given negative connotations. Discussions about the foreskin, both in the context of its pathological and physiological conditions have been going on for centuries, and include issues related to religion, hygiene, esthetics and tradition.

Literally, phimosis, from Greek φιμοσισ (muzzling, i.e. from the shape not dysfunction), is the condition of limited retraction or complete non-retraction of the foreskin over the glans penis. The inability to expose the glans penis in boys after birth is a physiological phenomenon. The foreskin should become entirely retractable until puberty and there is no concern to force it earlier [1]. In some cases, when retraction of the foreskin is impossible (or the inability to retract occurs after physiological phimosis subsides) despite the passage of time, the development of pathological phimosis is indicated [2,3]. Then phimosis is classified as primary or secondary. There are no signs of scarring in primary phimosis. Secondary phimosis presents scarring, residual discharge or inflammation. In 2002, researchers from London recognised that secondary phimosis corresponds to lichen sclerosus et atrophicus, better known as balanitis xerotica obliterans (BXO) [1], constituting it the only absolute indication for circumcision in boys.

The disease process affects the prepuce, glans and occasionally the urethra. Chronic irritation of the mucous membrane, glans penis inflammation (balanitis), and foreskin inflammation (posthitis) occur as a result of the secretions and exfoliated epithelium accumulation between the glans and foreskin [4,5,6]. The chronic inflammation that develops this way, which is referred to as constricting inflammation of the glans and foreskin, leads to remodelling of the tissue matrix. The increased production of pro-inflammatory cytokines and the influx of chemokines force fibroblasts to become fibrotic at the inflammed site [4,7]. Early BXO symptoms in the physical examination of the penis and foreskin are hypopigmentation, erythematous spots, and purple-white plaques. Fully developed BXO is manifested by white plaques and sclerosus within the glans, penis, and foreskin [8,9]. Histological examination results are characterised by the following features – hyperkeratosis with follicular plugging, atrophy of the stratum spinosum malpighi with hydropic degeneration of the basal cells, lymphoedema, hyalinosis, the homogenisation of collagen in the dermis, and an associated band-like chronic inflammatory cell infiltrate. The disorder typically presents with irritation, local infection, dysuria, bleeding, secondary non-retractability of the foreskin or a deteriorating urinary stream. On rare occasions, it can progress to the point of presenting with acute urinary retention or secondary diurnal or nocturnal enuresis resulting from chronic outflow obstruction. This reveals two important considerations: circumcision should be performed only in BXO cases, and patients require follow-up monitoring due to the risk of urethral stricture.

As the data from the literature are limited and unequivocal, the authors aim to study the prevalence of BXO. This study aims to assess the histopathological examination results of patients after surgical treatment of phimosis in order to ascertain the relationship between BXO and clinically diagnosed phimosis.

MATERIAL AND METHODS

This study analyzed the results of histopathological examinations in patients who underwent surgical methods of phimosis treatment in the period from January 2014 to March 2020 in the Department of Children's Developmental Defects Surgery and Traumatology in Zabrze. The inclusion criteria of the study were the surgical treatment of phimosis with the associated histopathological examination of the specimens collected during the surgery.

The data were collected prospectively and randomly – the decision regarding histopathological examination was not based on the surgeons' assessment and concerned all the patients operated by the surgical team participating in the study (of the two teams working in the department).

At the referral for surgery, all the patients were assigned grades III–V, according to the scale of foreskin retraction by Kikiros et al. [10] (Table I), while the decision regarding histopathological examination was routine, not based on clinical status.

Statistica 13.3 software (SoftPols, Krakow, Poland) was employed for statistical analysis. The V-Cramer coefficient, a variant of the chi-squared test for complex contingency tables, was used to calculate therelationship between the variables due to their nominal nature, taking values within 0–1, where 0 – stochastically independent variables, 1 – there is a functional relationship between the variables.

Based on the above-mentioned criteria, 106 subjects were enrolled in the study out of 350 who underwent surgical treatment of phimosis in the discussed period. All the patients were obviously boys, whose age range was 4–18 years (mean age 9.40 ± 3.82 years; median age – 9 years). In the group of patients included in the analysis, the following procedures were performed: circumcision, partial circumcision with preputioplasty, preputioplasty, and undercutting of the frenulum with preputioplasty.

Table I. Classification of phimosis severity according to foreskin retractability (based on [10])

0 (***no phimosis***) – full retraction, not tight behind glans, or easy retraction limited only by congenital adhesions to glans
1 – full retraction of foreskin, tight behind glans
2 – partial exposure of glans, prepuce (not congenital adhesions) limiting factor
3 – partial retraction, meatus just visible
4 – slight retraction, but some distance between tip and glans, i.e. neither meatus nor glans can be exposed
5 – absolutely no retraction

Table II. Distribution of surgical methods in group of patients who underwent histopathological evaluation after phimosis surgery

Operational method	Number of subjects	Proportion of subjects in whole group (%)	Mean age of subject ± SD (in years)
Circumcision	**84**/106	79.2	9.36 ± 3.74
Partial circumcision with preputioplasty	**3**/106	2.8	10.67 ± 4.93
Preputioplasty	**17**/106	16	9.00 ± 4.02
Undercutting of frenulum with preputioplasty	**2**/106	1.9	12.5 ± 6.36

RESULTS

There were 106 patient in whom a surgical procedure and histopathological examination were performed. The mean age of the patients was 9.41 ± 3.82 years. The share of individual methods of surgical treatment is shown in Table II. The age distribution of the patients with a histopathological diagnosis of BXO is shown in Figure 1. In the entire group of patients, the diagnosis of BXO (including BXO focal) was 59% (n = 63).

The vast majority (96%) of patients had histopathological conditions jeopardizing the integrity of the prepuces, confirming proper and health-related referral for surgery. Apart from BXO, there were 32% cases of an inflammatory process, and 3.7% fibrosis or keratosis. In one case (0.9%), the process was specific (bowenoid papulosis). In only 3.7% were there other conditions than inflammatory (naevus) or the skin was normal.

All the histopathological diagnoses in the studied group of subjects are presented in Table III.

The analysis showed that in the subjects diagnosed with BXO, the frequency of full circumcision was twice as high as in the patients with other histopathological diagnoses (56:28), while conservative surgery was performed much less frequently in this group (5:12). This is a statistically significant relationship ($p = 0.0108$), however, with a weak relationship, as indicated by the value of the V-Cramer coefficient (0.3246).

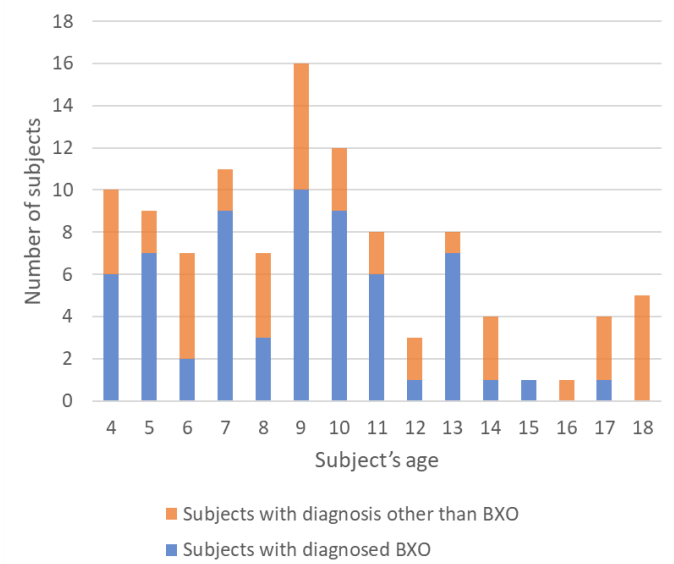

Fig. 1. Age distribution of subjects with histopathological diagnosis of balanitis xerotica obliterans (BXO).

Table III. Histopathological diagnoses in studied group of patients

Diagnosis	Number of patients	Proportion of patients in whole group (%)	Mean age of patients ± SD
Balanitis xerotica obliterans	52	49	8.65 ± 3.19
Balanitis xerotica obliterans focalis	11	10	9.8 ± 2.35
Inflammatio chronica	19	18	10.16 ± 4.37
Posthitis chronica	14	13	10.57 ± 4.77
Excessive inflammatory keratosis	1	1	6 ± nd
Naevus pigmentosus	1	1	6 ± n/a
Fibrosis and hyperemia	3	3	12 ± 4.32
Posthitis chronica cum fibrosis	1	1	9 ± n/a
Bowenoid papulosis	1	1	17 ± n/a
Normal skin	3	3	9.67 ± 6.66

There was no statistically significant correlation between the patient's age and the histopathological diagnosis (Spearman's test, V-Cramer test). In the study group, a lower percentage of BXO was found in patients over 14 years of age than in the entire group; nevertheless, the data is not significant due to the small number of patients (in this group, 11 circumcision operations were performed, 1 partial circumcision with preputioplasty, 1 undercutting of the frenulum with preputioplasty and 2 preputioplasty).

DISCUSSION

Topical glicocorticosteroid therapy is broadly used in phimosis therapy. Secondary phimosis or non-effective topical therapy in phimosis are indications for surgical treatment [3]. Surgical ways of treating phimosis give a chance to establish the aetiology of the disease by the histopathological examination of specimens collected during the surgery. The diagnosis of BXO should be confirmed by routine post-surgical specimen collection and histopathological examination.

The true prevalence of BXO is difficult to estimate and remains the subject of much research.

In this study, the prevalence of the histopathological features of BXO was found in 59% of patients who underwent surgery to release the foreskin. A similar incidence (50% and 52.6%, respectively) was described in an analysis conducted a year earlier by Ghidini et al. [11] and a decade earlier by Jayakumar et al. [12]. A prospective cohort study (Kiss et al. [6]) shows that this nonspecific inflammation is detected during histopathological examination after surgical removal of the foreskin or its retraction in about 10–40% of the samples taken.

Regarding the literature, we have noticed an increase in the proportion of diagnoses of constricting inflammation of the glans and foreskin as an etiological factor of phimosis, however, as some studies report, BXO is not the only etiological factor of phimosis [1]. Our study determined the proportion of diagnoses other

than BXO in patients with clinically diagnosed phimosis at the level of 41%.

In a review, Celis et al. [13] estimated the global incidence of BXO in circumcised boys at 35%. In the study group, the incidence of BXO was higher than that indicated, among others, by Boksh and Patwardhan [14], Bochove-Overgaauw et al. [15], Yardley et al. [16] or Kiss et al. [6] – respectively, 5.5%, 27%, 34.5%, 40% in the population of boys under 16. The same was with most of the cases in our study.

The conclusions from the cited literature and this analysis suggest that the incidence of BXO may be higher than previously thought, but the qualification for surgery in the cited studies could also include milder cases of phimosis, thus making the aforementioned studies incomparable.

It is worth mentioning that our study group contains only those patients with grade III–V according to Kikiros and almost all of them with other histopathological conditions, most of them being chronical inflammation (the overall number being 96%). Shankar and Rickwood [17] in a population of boys aged 5 to 14 years reported BXO in 84% of patients, which exceeds the value determined in this study.

The rising incidence of BXO diagnoses may be attributed to an increased submission of intraoperative samples for histopathological examination and a more reduced tendency to refer patients for surgery. Phimosis without the clinical symptoms of BXO in our studies often gave a positive result with the features of this condition on histopathological examination.

It is necessary to note that post-operative histopathological examination is not common (not all centres perform it routinely), and only taking a circumcised foreskin for examination after the surgical treatment of phimosis allows the diagnosis of BXO, which in turn may contribute to the possible prolonged follow up and/or use of additional therapy with topical glicocorticosteroids or topical immunosuppressants.

Such therapy protects against the possible recurrence of inflammations and their consequences (urinary retention, urethral stricture) [15], as well as against a persistent medical condition involving the glans and urethra.

CONCLUSIONS

Although available publications indicate that the incidence of BXO in patients undergoing surgical methods of treating phimosis is 5.5–84% [1,11,12, 14,15,16,17], most of them are in the medium of the range.

Differences in the incidence of BXO in the cited studies suggest that the available evidence is not sufficient to accurately determine the incidence of the disease and that the subject needs to be developed for further research and analysis, as well as standardization of the criteria of referring for surgery.

However, we can conclude that BXO is the most common cause of visible scarring of the prepuce, though it is not the sole indication for surgery of secondary phimosis. The other common causes of phimosis include, e.g. chronic inflammation and posthitis.

Statements

The authors have no competing interests to declare. The work was not sponsored or funded by any external source or institution. The ethical committee of the affiliated institution was informed about the research and stated that it does not require ethical committee approval.

Author's contribution

Study design – W. Korlacki, A. Grabowski
Data collection – W. Korlacki, A. Grabowski
Data interpretation – P. Pobudejski
Statistical analysis – M. Migas
Manuscript preparation – P. Pobudejski, B. Meier, M. Migas
Literature research – P. Pobudejski, B. Meier, M. Migas

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