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ORIGINAL ARTICLE

Prevalence and surgical management of concurrent adult acquired buried penis and urethral stricture disease

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Abstract

Purpose To describe the prevalence and surgical management of coexistent adult acquired buried penis (AABP) and urethral stricture disease. AABP patients often have urinary dribbling with resultant chronic local moisture, infection, and inflammation that combine to cause urethral stricture disease. To date, no screening or surgical management algorithms have been described.

Methods A multi-institutional retrospective study was conducted of the surgical management strategies for patients with concurrent AABP and urethral stricture disease from 2010 to 2017. AABP patient demographics, physical exam findings, and comorbidities were compared between those with and without stricture disease to suggest those that would selectively benefit from screening for stricture disease.

Results Of the 42 patients surgically managed for AABP, 13 had urethral stricture disease (31.0%). Stricture location was universal in the anterior urethra. Sixty-one percent ($n=8$) of strictures were 6 cm or longer and managed prior to AABP repair with Kulkarni urethroplasty. Patients with urethral stricture disease were significantly more likely to have clinically diagnosed lichen sclerosus ($p=0.00019$). There was no significant difference in BMI, age, or comorbidities between patients with and without urethral stricture disease.

Conclusions Extensive anterior urethral stricture is common in patients with AABP. Clinical characteristics cannot predict stricture presence except possibly the presence of lichen sclerosus. Definitive stricture surgical options include extensive Johanson Urethroplasty or Kulkarni Urethroplasty. Kulkarni Urethroplasty prior to AABP repair has the benefits of a single-stage repair, good cosmetic outcome with meatal voiding, and dorsal graft placement to allow safe degloving of the penis in the subsequent AABP repair.

Keywords Adult acquired buried penis · Urethral stricture · Lichen sclerosus · Panurethral stricture disease · Kulkarni urethroplasty

Introduction

Adult acquired buried penis (AABP) is a morbid condition that is rising in incidence due to the obesity epidemic, although the exact incidence is unknown. The most frequent etiology is obesity, but the condition can also be a result of post-circumcision cicatrix formation or primary genital lymphedema. Regardless of etiology, affected patients very frequently have urinary dribbling that causes chronic local moisture, infection, and inflammation [1, 2]. Lower urinary tract symptoms (LUTS) are nearly universally present in this population either solely due to the inflammatory microenvironment or in tandem with concurrent lichen sclerosus and urethral stricture disease [3]. Differentiating LUTS due to penile burying and inflammation from urethral stricture disease is paramount because surgical repair of AABP prior to surgical stricture management can limit treatment options.

The urethral stricture disease associated with lichen sclerosus is often severe with lengthy strictures that require extensive repair. Surgical management options include Kulkarni type urethroplasty, extended first- and second-stage Johanson urethroplasty or perineal

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urethrostomy [4–6]. AABP repair associated with morbid obesity often requires split-thickness skin grafting (STSG) due to penile skin fibrosis as a result of burying [1, 2]. The presence of STSG limits surgical options for long-segment stricture disease if AABP repair is pursued prior to stricture management. For example, penile inversion during Kulkarni urethroplasty after AABP repair is complicated by the presence of STSG. The STSG would need to be divided longitudinally in a Johanson approach and there would be a lack of dartos fascia to create a layered repair risking graft loss [7]. These surgical considerations must be taken into account prior to AABP repair. It is especially unfortunate if a surgeon who is unaware of a urethral stricture inadvertently attributes a patient's voiding symptoms to those commonly seen with a buried phallus and thus limits a patient's surgical options.

We hypothesize that there are a high number of patients with urethral stricture disease within the AABP population. Herein, we report a multi-institutional review of AABP patients to define the prevalence of concurrent stricture disease and suggest clinical characteristics that can imply their presence. Based on this cohort, we suggest an algorithm for workup and management of patients with LUTS and AABP to optimally detect and sequentially surgically manage AABP and urethral stricture disease.

Materials and methods

After institutional review board approval at two institutions, patients surgically managed for AABP from 2010 to 2017 were retrospectively reviewed. Patients with AABP were screened for urethral stricture disease based on surgeon suspicion of urethral stricture disease or difficulty with catheter placement. Screening included cystoscopy or retrograde urethrogram (RUG).

Patient demographic collection included age, body mass index BMI, and comorbidities including diabetes, hypertension, and obstructive sleep apnea. Urethral stricture-specific details including the presence or absence of stricture disease, stricture location and length, incidence of lichen sclerosus based on clinical exam, and urethral stricture surgical management specifics were collected. Special attention was given to the order of stricture surgical repair in regard to their AABP repair (stricture requiring treatment prior to AABP versus concordant repair or adjuvant stricture repair) and the surgical approach required for stricture repair (Johanson, augmented buccal onlay/inlay, anastomotic, panurethral stricture disease with Kulkarni type urethroplasty, etc.). Using a *t* test, comparisons were made between AABP patients with and without urethral stricture disease.

Results

Over the study period between 2010 and 2017, 42 patients were surgically managed for adult acquired buried penis. Thirteen patients (31.0%) were found to have urethral stricture disease requiring surgical management. Thirty patients (71.4%) required split-thickness skin graft during AABP repair including all (13 of 13) patients who were also surgically managed for urethral stricture disease. One of the 13 patients who did not receive skin grafting at time of AABP repair had reburying and obstructive symptoms requiring multiple endoscopic procedures for stricture disease. This patient ultimately underwent a second AABP repair with STSG and has been free of obstructive urinary symptoms at 34 months.

Stricture location was universally in the anterior urethra. The majority of patients with urethral stricture were identified preoperatively based on surgeon suspicion with plans for cystoscopy, retrograde urethrogram (RUG) either prior to or at time of AABP repair. Mean stricture length for the cohort was 5.8 ± 3.8 cm (SD). Sixty-one percent ($n=8$) of strictures were 6 cm or longer.

Of the thirteen patients with stricture disease, seven were treated preoperatively, five were treated simultaneously and one was treated postoperatively. Patients with long-segment anterior urethral stricture disease were primarily managed with Kulkarni type urethroplasty prior to AABP repair ($n=6$). For the seven patients not treated with Kulkarni urethroplasty, including those with shorter anterior urethral strictures and meatal strictures, management included a combination of concurrent first-stage Johanson ($n=4$) or intraoperative urethral dilation ($n=3$) with AABP repair (Table 1). 11 patients (85%) required no additional treatment for stricture disease. Two patients demonstrated lower urinary symptoms after their AABP and stricture management, but did not require additional dilations or endoscopic procedures.

There was no significant difference in BMI, age, or comorbidities, between patients with and without urethral stricture disease (Table 2). Patients with urethral stricture disease were more likely to have lichen sclerosus based on clinical exam ($p=0.00019$).

The preoperative knowledge of the presence and severity of urethral stricture disease changed management in 10 of 42 patients managed for AABP (23.4%). Preferred surgical management of the patient's urethral stricture disease would have been deleteriously affected by AABP repair prior to urethral stricture repair in this subset.

Table 1 Adult acquired buried penis stricture length, location, and surgical management

Stricture location	Stricture length (cm)	Surgical management	Stricture management (compared to timing of AABP repair)
AABP–urethral stricture characteristics and management			
Penile	6	Kulkarni	Preop
Penile/meatal	12	Dilation	Preop
Penile	7	Kulkarni	Preop
Penile	3	1st stage Johanson	Simultaneous
Penile/meatal	13	Kulkarni	Preop
Distal penile	2	1st stage Johanson	Postop
Distal penile	2	Dilation	Simultaneous
Penile/bulbar	6	Kulkarni	Preop
Distal penile	2	1st stage Johanson	Simultaneous
Meatal	2	Dilation	Simultaneous
Penile	10	Kulkarni	Preop
Penile	6	Kulkarni	Preop
Penile/meatal	5	1st stage Johanson	Simultaneous

Table 2 AABP patient demographics, comorbidities, and physical exam findings with and without urethral stricture disease

	AABP	AABP and stricture	*p*
Adult acquired buried penis			
Age (years)	53.7	49.7	0.21
BMI (Kg/m$^{2)}$	43.6	42.6	0.34
Lichen Sclerosus (%)	17.2	71.4	0.00019
Comorbidities (%)			
OSA	38	38.5	0.49
DM	51.7	46.2	0.37
HTN	68.9	46.2	0.095

Discussion

Surgery for the treatment of adult acquired buried penis results in substantial improvements in quality of life. AABP is associated with not only urinary and sexual morbidity but also substantial psychological distress. Affected patients often suffer from depression and anxiety. Surgical repair improves urinary and sexual outcomes in addition to body image and mood [8–11].

Additionally, it is increasingly recognized that AABP patients frequently develop lichen sclerosus as a result of chronic moisture and inflammation from urinary dribbling with subsequent urethral stricture development [3]. As the incidence of buried penis increases with the growing obesity epidemic, the frequency with which surgical repair is offered will almost certainly follow suit. Given the frequency with which AABP and urethral stricture occur together, it is prudent to develop a systematic algorithm including up-front screening for stricture disease in the AABP population. In the study by Theisen et al. 2018, the majority patients undergoing AABP repair reported significant improvements in urinary symptom scores postoperatively suggesting that a validated voiding symptom questionnaire could be used as part of screening algorithm [12]. It is especially important to identify stricture disease prior to AABP surgical intervention to preserve stricture surgical treatment options. AABP frequently necessitates split-thickness skin grafting, which complicates if not eliminates perineal penile invagination techniques for buccal mucosa onlay procedures and a multi-layered closure in a two-stage Johanson approach. All thirteen of our patients with AABP and stricture disease ultimately required STSG.

We suggest screening all patients with AABP for urethral stricture disease using the validated voiding symptom questionnaire, such as the AUA-SI, with subsequent retrograde urethrogram or cystoscopy if stricture disease is implied. Retrograde urethrogram has the benefit of defining the length and severity of urethral stricture if present but can often be technically difficult with a completely buried phallus. Semirigid urethroscopy is a useful addition to the armamentarium to define urethral stricture disease in AABP. It allows negotiation of the skin tract in a completely buried phallus and is able to traverse most meatal and fossa strictures to define the length of the stricture disease (Fig. 1).

Meatal and distal penile strictures are relatively easily managed at the time of AABP repair with either a first-stage Johanson urethroplasty or Nikolavsky ventral buccal inlay approach. Longer segment strictures often necessitate perineal urethrostomy or perineal approach panurethral dorsal onlay urethroplasty (Kulkarni urethroplasty). Based on the existing literature about urethral stricture recurrence after

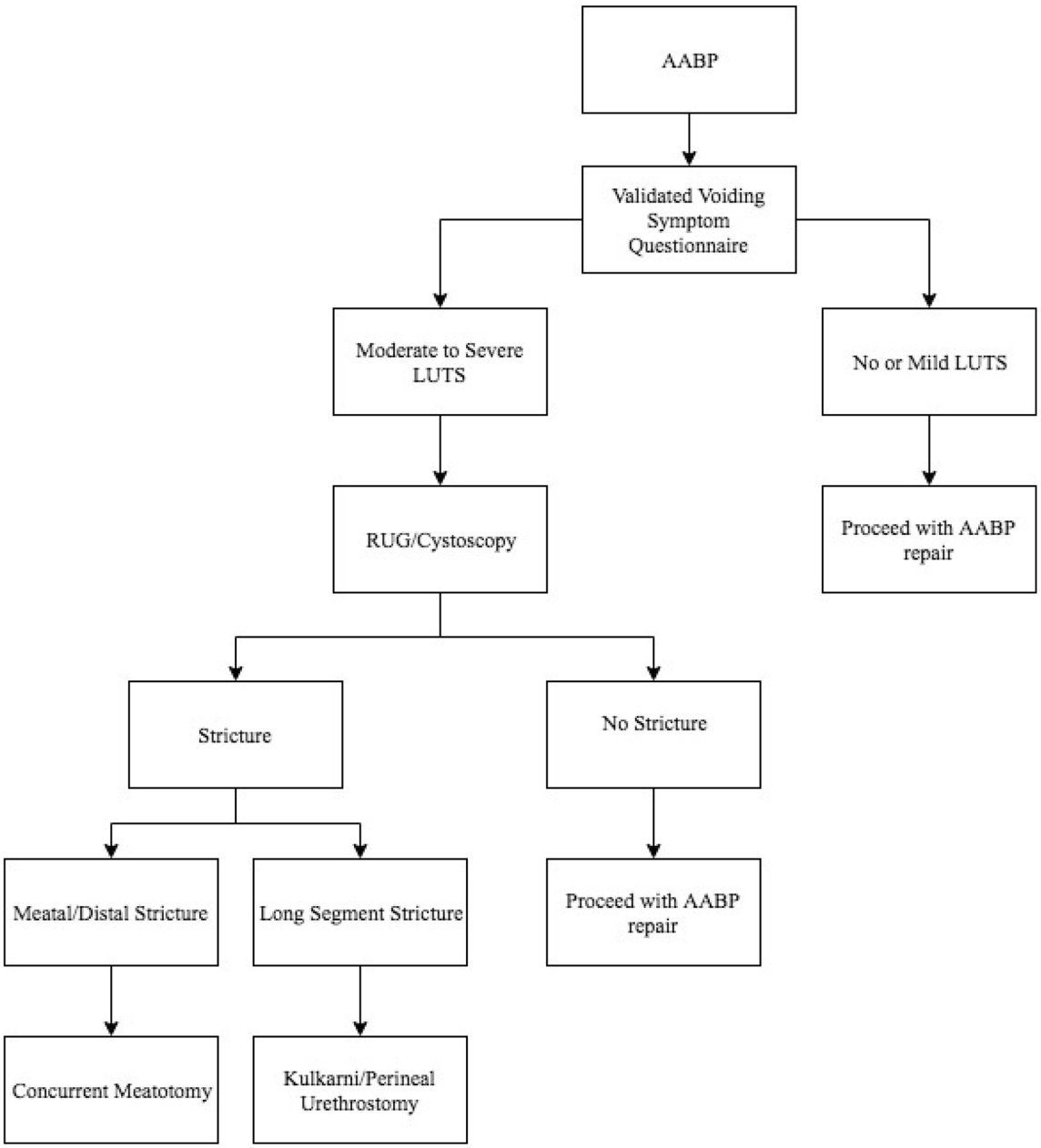

Fig. 1 Suggested patient clinical evaluation and surgical management algorithm for AABP with concurrent urethral stricture disease

urethroplasty, we would recommend a minimum 6-month delay between initial stricture management and AABP repair, if the procedures are not performed simultaneously [13]. It is not uncommon to have young patients with AABP given rising childhood obesity rates and in general patients functional enough to consider AABP repair who desire meatal voiding, so Kulkarni urethroplasty prior to AABP repair is a preferred approach over urinary diversion with perineal urethrostomy.

Limitations of our study include the retrospective nature, dependence on surgeon suspicion or difficulty with catheter placement for identification of stricture, dependence on clinical exam (not pathology) for diagnosis of lichen sclerosus, lack of preoperative and postoperative validated voiding screening questionnaires and a relatively small sample size.

Conclusions

Adult acquired buried penis and long-segment anterior urethral stricture disease are common concurrent pathologies with 30% of our cohort having urethral strictures which is

consistent with the incidence described in Liaw et al. The chronic moisture and high-pressure voiding due to a buried phallus leads to an inflammation of the periurethral glands and subsequent lichen sclerosus and urethral stricture disease [3]. Identification of stricture, location, and length is necessary prior to AABP repair to appropriately plan a staged surgical repair. We would advocate more aggressive routine screening for patients with AABP to identify patients with concurrent stricture disease using a validated voiding screening questionnaire as an initial screening tool with a low threshold to perform urethral calibration, RUG and cystoscopy if patients have moderate to severe symptoms of LUTS.

Meatal, fossa, and distal penile strictures are easily managed at the time of AABP repair with short first-stage Johanson urethroplasty. Bulbar urethral stricture repair can be treated via the perineum with anastomotic or augmented urethroplasty. Long-segment anterior urethral stricture disease necessitates a more thoughtful approach. Johanson urethroplasty is not technically possible or advisable prior to AABP repair with long-segment anterior strictures. Postoperatively from AABP repair second-stage Johanson is complicated by the loss of dartos to create a layered repair and the frequent presence of split-thickness skin graft. Kulkarni urethroplasty for long-segment anterior urethral stricture disease prior to AABP repair has the benefits of a single-stage repair, durable stricture results, good cosmetic outcome with meatal voiding, and dorsal graft placement to allow safe degloving of the penis in the subsequent AABP repair.

Author contributions TWF: project development, data analysis, and manuscript writing. KP: manuscript writing/editing and data collection. KMT: data collection. AT: data collection. FB: project development. PJR: project development

Compliance with ethical standards

Conflict of interest The authors state they have no financial or other conflicts of interest to disclose.

Ethical approval This retrospective study underwent multi-institutional IRB approval and involved minimal risk to the subjects. Care was taken to protect patients' health information and confidentiality of collected data. For this type of study formal consent is not required.

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