Perineal Cyst in Transgender Men: A Rare Complication Following Gender Affirming Surgery — A Case Series and Literature Overview

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ABSTRACT

Introduction: Genital gender affirming surgery (gGAS) is usually the final stage in the medical transition for transgender men and consists of creating a neophallus and neo-scrotum, with or without urethral lengthening (UL). To reduce the complication risks of UL, a mandatory colpectomy is performed prior to UL. Colpectomy is considered a complex surgery, which may lead to various perioperative complications. There are few long-term complications reported.

Aim: To describe the clinical presentation and management of 3 consecutive transgender men presenting with a perineal cyst following gGAS.

Methods: After obtaining informed consent all clinical data was collected, including medical history, current symptoms, imaging, as well as surgery and histological outcomes. Furthermore, a literature search was performed.

Main outcome measure: To hypothesize the aetiology of the perineal cyst based on current published literature.

Results: Three otherwise healthy transgender men, ages 26—46 with a similar medical history, presented with a perineal cyst several months or years following colpectomy and gGAS with UL. All patients underwent surgery to remove the cyst. Several theories regarding aetiology of this perineal cyst are discussed in this report.

Conclusion: There remain several gaps in our knowledge regarding the aetiology and management of this perineal cyst. Therefore, further research is necessary. **Asseler JD, Ronkes BL, Groenman FA, et al. Perineal Cyst in Transgender Men: A Rare Complication Following Gender Affirming Surgery — A Case Series and Literature Overview. J Sex Med 2021;9:100415.**

Key Words: Transgender; Female-to-male transgender; Colpectomy; Vaginectomy; Gender affirming surgery; Urethral lengthening surgery; Perineal cyst; Vaginal remnant

INTRODUCTION

Most transgender persons feel dysphoria towards their internal and external genitalia.[1] Genital gender affirming surgery (gGAS) is usually the final stage in their medical transition. In transgender men (FtM; female-to-male transgender), gGAS consists of creating a neo-scrotum and a neophallus with or without urethral lengthening (UL). A majority of transgender men wish to achieve voiding whilst standing following Ggas.[2] To achieve this, UL is necessary. Common complications in UL include urethra strictures, obstructions or fistula's. To reduce the risk of fistula formation, a colpectomy, or vaginectomy, prior to UL is mandatory in our centre.[3] Following colpectomy, in a second surgery, phalloplasty or metoidioplasty and UL are executed. The 2 colpectomy techniques used in the Centre of Expertise on Gender Dysphoria in Amsterdam, are the vaginal approach and the robot assisted laparoscopic approach. A detailed description of our vaginal and robot assisted laparoscopic surgery techniques was previously published by Nikkels et al[4] and Groenman et al,[5] respectively. Contrary to common electrocautery and/or ablation

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Table 1. Overview of clinical data of the patients

	Patient		
	No. 1	No. 2	No. 3
Age	28	46	26
BMI	23	23	23
WHO performance status	0	0	0
Smoker	No	No	No
Surgical history	Mastectomy, total laparoscopic hysterectomy, colpectomy, phalloplasty with urethral lengthening	Tonsillectomy, mastectomy, metoidioplasty, total laparoscopic hysterectomy and bilateral salpingo-oophorectomy, colpectomy, phalloplasty with urethral lengthening, testicular prosthesis	Mastectomy, total laparoscopic hysterectomy and bilateral salpingo-oophorectomy, colpectomy, phalloplasty with urethral lengthening, testicular prosthesis
Medical history	Gender dysphoria, depression, gastroesophageal reflux disease	Gender dysphoria, asthma	Gender dysphoria
Current medication	Nebido®[Bayer, Germany], fluoxetine, omeprazole	Androgel®[Besins international, Belgium], salbutamol, formoterol/beclometasone, prednisolone	Sustanon®[Aspen Pharma Trading Limited, Ireland]
Colpectomy technique used	Robot assisted approach	Vaginal approach	Vaginal approach
Time colpectomy to presentation	3 years	7 years	5 years
Time colpectomy to phalloplasty with UL	2 years	2 years	1 year
Time phalloplasty to presentation	9 months	5 years	4 years
Time last urethroplasty to presentation – reason urethroplasty	6 months – urethroscrotal fistula	4 years – urethra stricture	1 year – meatal stenosis
Maximum size of defect on magnetic resonance imaging (MRI)	1.8 cm	10.1 cm	5 cm

strategies, both approaches used in our centre dissect the complete vaginal epithelium as thinly and precisely as possible in a submucosal plane to prevent nerve injury to adjacent structures, to prevent bleeding from the perivaginal plexus and to prevent fistula to bladder, urethra or rectum. After removal of the entire vaginal wall, the vaginal apex is closed by suturing the remnants of the rectovaginal septum and endopelvic fascia of the vesicovaginal space together. Colpectomy is considered a complex surgery, which may lead to various perioperative complications like haemorrhage or bladder and bowel injuries.[4] There are few long-term complications reported. This case series describes 3 transgender men who, years later, presented with a perineal cyst following colpectomy and gGAS with UL.

CASES

Three transgender men presented with a perineal swelling. Their ages ranged from 26 to 46 years. Symptoms at presentation consisted of perineal pressure, bulging mass during Valsalva manoeuvre or spontaneous and aesthetic discomfort. There were no signs of infection during physical examination and no voiding complaints were mentioned. All men were receiving exogenous testosterone treatment and were otherwise healthy. Their g(GAS) history consisted of a mastectomy, hysterectomy, colpectomy and phalloplasty with UL. All patients underwent previous urethroplasty, due to complications of their UL.

One patient combined his previous hysterectomy with a colpectomy, using the robot assisted laparoscopic approach. Two patients underwent a vaginal colpectomy in a separate procedure after their hysterectomy. The time between colpectomy and the occurrence of the swelling varied between 3 and 7 years. One to 2 years following colpectomy, all patients underwent gGAS with UL. The time between gGAS and the occurrence of the swelling varied between 9 months and 5 years. In this period, all patients received additional urethroplasty to treat complications from the UL (ie, urethra fistulas and/or strictures). For an overview of the patients clinical data, see Table 1.

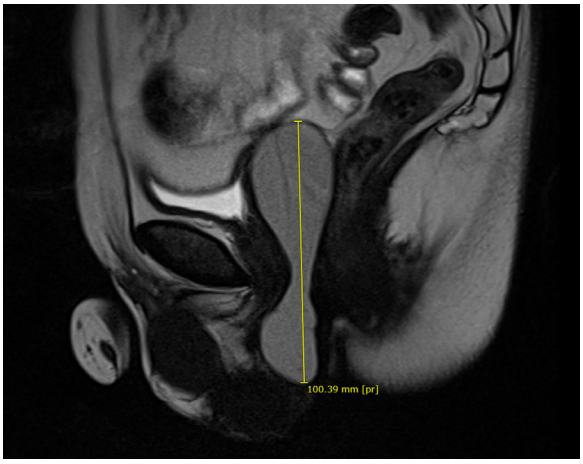

Figure 1. Magnetic resonance imaging scan in T2 setting shows an extensive fluid filled cavity of approximately 10 cm.

Magnetic resonance imaging (MRI) was performed to determine size and relation to adjacent structures. The MRI confirmed a local fluid collection, or collections, in the perineal midline. In 2 patients the fluid collection was located dorsally from the urethra. In 1 patient complex, multiple fluid collections expanding from the midline were described. The maximal size varied between 1.8 cm and 10.2 cm (Figure 1). There were no fistulas, to or from the cyst, found on MRI.

All patients underwent surgical removal of the cyst via longitudinal incision in the perineal skin (Figure 2). After blunt and sharp dissection of the caudal part of the cyst, the cyst was opened and the fluid inside drained. The fluid was described as clear-green and mucinous in 2 patients and yellow-brown and puss-like in one patient. Thereafter, the cyst wall was carefully removed due to proximity of adjacent bowels, bladder etc. In 2 patients (patient 1 and 2) a urethral fistula to the cyst was present, which was not visible on the prior MRI. The fistulas were treated in the same procedure. The skin was closed using sutures and a perineal drain was left behind. There was no peri- or postoperative complications reported. One patient has since received a (temporary) perineostoma due to persistent fistula formation (patient 1). The other patients reported no micturition complaints.

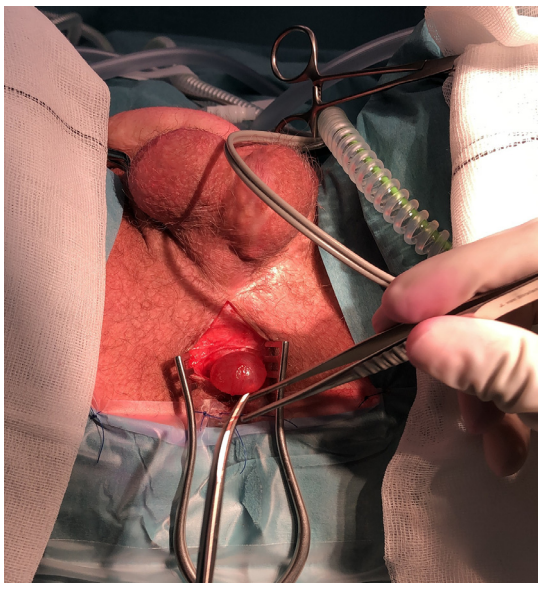

Figure 2. Perioperative image of the protruding cyst after longitudinal incision in the perineal skin.

Histological examination showed all cyst walls were (partially) covered in squamous epithelium, similar to vaginal epithelium and showed smooth muscle tissue as well. There were signs of moderate to severe inflammation and granulation tissue. In 1 patient (patient 1) a Bartholin gland was recognised, indicating a Bartholin cyst or abscess. There were no signs of urothelium or malignancy.

DISCUSSION

At the time of writing, we performed 178 robot assisted colpectomies and 184 vaginal colpectomies in the Centre of Expertise on Gender Dysphoria in Amsterdam. Indicating that perineal cyst formation following gGAS is rare. It might be possible there are more patients with a perineal cyst, but who have not (yet) become symptomatic.

In literature similar findings are mentioned. Most notably the studies by Nikolavsky et al.[6,7] The authors describe a fluid filled "vaginal cavity" following colpectomy and UL in over half of their patients presenting with a stricture in their neourethra. They hypothesize the distal obstruction caused by the urethra stricture, leads to pressurised urine to find its way to the previous location of the vaginal cavity. When performing corrective urethroplasty, the "vaginal cavity" was excised and send for histological examination, showing normal vaginal epithelium. No theory was provided on how this vaginal epithelium remained or was restored on the previous location of the vaginal cavity. There was limited information provided regarding patient data, surgery techniques and imaging in both studies. Furthermore, none of our patients presented with a urethra stricture.

Similar to Nikolavsky, a study by Dy et al,[8] showed a "vaginal remnant" in 47% of patients. The authors hypothesize the incomplete removal of vaginal epithelium during colpectomy, may lead to secretion of set epithelium whilst the introitus is already surgically closed. The accumulation of these secretions may result in a mucocèle and promote re-epithelialization of that cavity. In contrast to Nikolavsky et al, Dy et al describes an often simultaneous occurrence of fistula's as a result of this "vaginal remnant" instead of being the cause.

Various colpectomy techniques may affect the chance of developing a perineal cyst following gGAS. Contrary to the colpectomy technique used in our centre where we dissect the entire vaginal epithelium, other centres may use an ablative procedure with electrocautery to create scarring and closure of the vaginal

cavity.[9,10] Studies by Nikolavsky and Dy do not clarify which colpectomy technique was used, however this might explain the difference in incidence in our population compared to the percentages mentioned above.

Two other studies by Stojanovic et al[11] and Al-Tamimi et al[3] mention the presence of a "perineal vaginal mucosa cyst" or a "persistent vaginal cavity" in 9 of 473 and 2 of 473 patients respectively. There is no further data described regarding this complication, only that they all required surgical removal. Additionally, one case report by Young et al[12] described a 45-year old trans-man with a urethral stricture, a urethrocutaneous fistula and a "vaginal remnant" after a colpectomy and gGAS. In this article, no further details are described.

Another origin theory, is the development of a bartholin cyst after the complete closure of the vagina during gGAS. The bartholin glands are mucus secreting glands, positioned in the lower left and right section of the introitus.[13] When creating the neo-scrotum during gGAS, the introitus is closed. Perhaps enclosing the exit of these bartholin glands, resulting in an accumulation of mucus, inflammation, re-epithelization and thus cyst development. Since the bartholin glands are not identifiable upon palpation, there is no strategy to avoid or remove them during colpectomy or gGAS. This theory would suit with our histological findings in 1 patient (patient 1). However, if this theory is legitimate, it is peculiar we do not see this complication more often. In literature, only 1 case report describes the occurrence of a bartholin cyst in a transgender man.[14] In this study however, it is unclear if the patient also underwent colpectomy during his gGAS.

Based on the above described theories, we cannot find an all-encompassing theory for all 3 patients. Furthermore, in our third patient none of the separate theories seem applicable.

Not only the complete origin of the cyst remains unclear, when the cyst develops is unclear as well. In this case series, there are only 3 patients included, and their timeline varied significantly. Making it difficult to draw conclusions. An MRI was performed in 1 of our 3 patients due to persistent lower abdominal pain in between colpectomy and gGAS, showing no signs of a perineal cyst. No further imaging was performed in our patients that could suggest time of development.

No studies found, provided data on when the cyst occurred in relation to other clinical events, nor was there theorized how quickly the cyst might have developed. One prospective study we found however, described a mean follow-up of 44 months in which 9 perineal cysts developed.[11]

In our centre, colpectomy is mandatory before gGAS with UL in order to reduce post-operative urethral fistula formation.[3] This might differ in other centers where different consecution protocols are followed. Unfortunately, only 3 studies describing perineal cysts, describes their consecution protocol. Studies by Stojanovic et al[11] and the case report by Young et al[12] describe a 1-stage surgery, where colpectomy, phalloplasty and UL are all performed in 1 surgery. And the study bij Al-Tamini et al[3] describes a protocol where colpectomy is performed at least 3 months prior to phalloplasty and UL. It may be possible the healing time of the colpectomy wound may decrease the chance of developing a perineal cyst.

To gain information regarding the development and prevalence of the cyst, we suggest performing a physical examination and imaging in asymptomatic patients after their colpectomy on multiple occasions. Least strenuous for the patient would be to perform a perineal ultrasound before surgery, after the patient is under general anaesthesia. Suitable surgeries would be phalloplasty with UL or urethroplasty. This strategy for exploration and treatment (ie, surgical removal of a perineal cyst) during urethroplasty has been recommended by Scahrdein et al[15] based on the findings of Nikolavsky et al.[6,7] However, this standard exploration is not yet standard practice for asymptomatic patients at our centre.

In our literature search we found varying terminology to describe this long-term complication following colpectomy and gGAS. We recommend not to use the term "vaginal remnant" when describing this complication. As it may be inaccurate as well as insensitive and trigger gender dysphoria in the transgender male patient.

CONCLUSION

The long term complication of a perineal cyst formation following gGAS is a rare occurrence in our gGAS population and when it does, the experienced complaints are mild. In all cases however, surgical removal of the cyst was necessary. There remain several gaps in our knowledge regarding the aetiology and management of this cyst. Therefore, further research is necessary.

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Conceptualization, J.A., B.R., F.G., R.L., G.P. and N.M.; Writing – original draft, J.A.; writing – review & editing, J.A.; Supervision, N.M.

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