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Benchmarks for Partial Segmental Thrombosis of the Corpus Cavernosum: A Case Report and Review of the Literature

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Abstract

Introduction: Partial segmental thrombosis of the corpus cavernosum (PSTCC) is an uncommon clinical condition; it is known in the literature as partial priapism which chiefly affects young men; the underlying reasons and its pathogenesis are inadequately understood.

Case report: A 26-year-old male presented with a perineal swelling and pain of 11 hours duration, which had started after 10-hour flight, and a dull aching pain with insidious onset. The patient denied a history of excessive sexual activities, drug abuse, or trauma with unremarkable past medical history.

Aims and methods: With deficiency of curd data for PSTCC, only case reports without precisely case numbers; therefore all existing case reports, whether abstracts or full text, under the title of partial segmental thrombosis of the corpus cavernosum or partial priapism have been screened and statistically analyzed.

Results and outcome: The total number of cases was 41 and chiefly affects young men: 63.4% of the patients are in the age group 20-40, with a mean age of 32.3 yrs. 10 cases 24.4% were treated surgically, 21 cases 51.2% were managed conservatively, while 3 cases 7.3% were treated by surgical aspiration ± injection or irrigation and 2 cases 4.9% were only follow up without treatment.

Conclusion: PSTCC is a rare non-emergency urological condition that usually has a good outcome. Conservative treatment with systemic anticoagulants has shown an excellent outcome and surgical treatment should be reserved for cases, if conservative management fails.

Keywords: Corpus cavernosum thrombosis; Penile thrombosis; Partial priapism; Priapism

Background

Partial segmental thrombosis of the corpus cavernosum (PSTCC) is an uncommon clinical condition, which is known in literature as partial priapism. It's chiefly affects young men; the underlying reasons and its pathogenesis are inadequately understood [1]. Unilateral proximal corporal thrombosis is the fundamental characteristic of PSTCC that can be confirmed clinically and visualized by means of radiological imaging [2]. The patients mainly present with perineal pain, a palpable perineal mass, with or without partial priapism. It is important to distinguish this condition from priapism (prolonged, painful penile erection > 4 hours) typically PSTCC manifests itself without an erection [3]. Diagnosis achieved by the clinical symptoms in combination with an imaging investigation, especially ultrasound and MRI [4]. In this paper we present a case of PSTCC in young man, who was successfully treated conservatively; moreover we will present a review of the literature under the search words "partial segmental thrombosis of the corpus cavernosum" or "partial priapism" and we will provide a descriptive meta-statistical analysis of these data by using an SPSS program.

Case Report

A 26-year- old male presented with perineal pain (5/10) and swelling of 11 hours duration. The pain started after a long flight of 10 hours as a dull aching pain with insidious onset. The patient denied any history of excessive sexual activity, drug abuse, hematuria, urinary tract infection, lower urinary tract symptoms, blood disease, trauma, or malignancy, with an altogether unremarkable past-medical history. The erection was intact (IIEF-5 23/25), Erection Hardness Score (EHS = 4). Physical examination revealed a firm tender mass about 7 cm in the right proximal corporal body, with flaccid distal corpora and glans and without signs of indurations or fluctuation. No signs of infection, malignancy, or hematologic abnormalities including sickle cell disease in laboratory investigation and negative urine toxicity screening.

An ultrasound of the perineum and penis proved presence of a mass in the right posterior cavernous body with slight compression of the left corpus cavernosum. Duplex-Doppler of the penis revealed an absence of flow in the mass and a decreased flow in the right cavernous artery compared to the left side.

MRI examination was performed subsequently; MRI imaging usually shows the thrombus at different types of signal intensity on T1 or T2 weighted images, depending on the stage of hemoglobin degradation.

In our case, the thrombus in the right corpus cavernosum measuring approximately 7 cm was depicted in a sub-acute phase, when the accumulation of methemoglobin results in increased signal intensity, mostly of the thrombus, on native T1 weighted images (Figure 1).

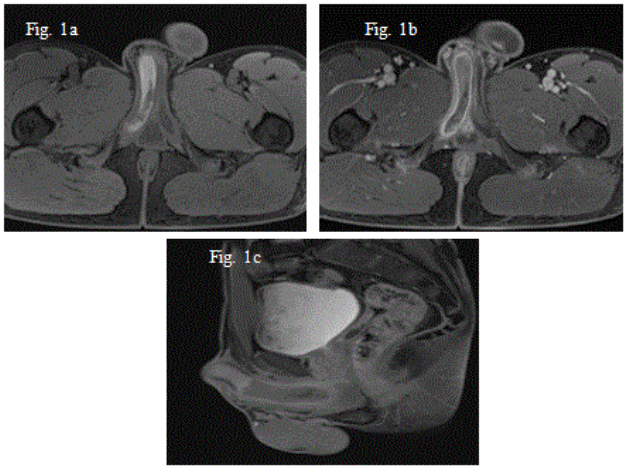

Figure 1: Axial fat-suppressed native T1 weighted image (1a) depicts the enlarged right corpus cavernosum with compression and displacement of the left cavernous body and for the most part a hyperintense thrombus signal, axial (1b) and sagittal (1c) fat-suppressed contrast-enhanced T1 weighted image shows the lack of enhancement of the thrombosed right side but a rim enhancement demonstrating a subtotal occlusion, Aera 1.5 Tesla MRI, Siemens, Erlangen, Germany).

After intravenous administration of a paramagnetic contrast agent, no enhancement was noted in the thrombus, but a hyperintense rim sign proving a subtotal occlusion of the cavernous body. The abnormal corpus cavernosum appears enlarged and compresses the displaced, non-affected contralateral cavernous body by shifting the midline.

We chose conservative management and after consulting a hematologist, the patient received weight-adapted low molecular heparin (Enoxaprin), beside analgesics and local measures. 4-weeks later, the patient reported preserved erectile function with no pain at all, the IIEF-5 score remained unchanged (23/25) and ultrasound showed the mass reduced in size. LMWH (1 mg/kg/day), 70 mg/day, continued for 3 months, followed by acetylsalicylic acid 100 mg once daily for another 3 months.

Material and Methods

In light of deficiency of curd or absolute data for PSTCC, all case reports in the literature, either abstracts or full text, under title partial segmental thrombosis of the corpus cavernosum, or partial priapism in literature were screened and evaluated in our analysis. The numbers of cases were summarized per decade, the mean age of the patients, clinical presentation, laterality, possible conceivable causes, strategies of management, complications and duration of follow-up were collected, evaluated and descriptive statistically analyzed.

Results and Outcomes

According to the literature, PSTCC is an uncommon clinical condition with aggregate quantities of cases 41 including our case (Table 1) [1-33], only five cases (12.5%) were diagnosed in the period between 1976 and 1984, seven cases (17.1%) between 1985 and 1994, ten cases (24.4%) between 1995 – 2004; in the last decade, 2005 and 2015, diagnostic frequency of PSTCC expanded and nineteen cases (46.3%) were recorded in this period. PSTCC chiefly affects young men, 63.4% of the patients are in the age group of 20-40, with a mean age of 32.3 ± 12.4 yrs.

All cases presented with acute painful mass in the perineum (proximal corpora), the duration of symptoms was less than one month in 91.2% of the cases, almost always unilateral: in 40 cases (97.6%) the symptoms were unilateral and only in one case (2.4%) it was bilateral described by Lewis et al. [21]. The persistence of the symptoms varied between 6 hours and 3 months.

Numerous causes were found that may instigate this condition, e.g. coitus or erection in 11 cases (26.8%), unremarkable causes in 11 cases (26.8%), cycling in 6 cases (14.6%), hematological disorders (i.e. spherocytosis [31], sickle cell anemia [21], hyperhomocysteinemia [6], protein C resistance [9], blood transfusion [15]) in 5 cases (12.2%), while alcohol or drugs abuse (marijuana, cocaine) may have been the cause in 3 cases (7.3%); in 2 cases (4.9%) the condition was associated with long distance flights and one case each (2.6%) was caused by Sildenafil [14] and Tamsulosin [18]. One or more of these elements may consider being the cause of PSTCC (Table 1).

No.	Year	Investigator	Age (yr)	Duration	Preceding Activity Other Factors	Treatment	Follow-up	Potency & Complication
1	1976	Gottesman [12]	34	4 days	Prolonged intercourse 10 prior events in 10 years	Surgical exploration with incision of corpora	1 mo	Yes
2	1976	Hillis and Weems [13]	24	11 days	Idiopathic	Surgical exploration and removal of transverse membrane	6 wk	Difficulty with full Erection
3	1980	Johnson and Corriers [17]	34	7 days	Prolonged intercourse	Surgical exploration and removal of transverse membrane	18 mo	Yes

4	1980	Llado [22]	23	14 days	Idiopathic Daily bicycling 2.5y FUO, anemia, prostatitis	Surgical irrigation, then cavernosalspongioum shunt	3 mo	Yes
5	1981	Chary et al. [8]	46	3 days	Idiopathic	Analgesics	3 mo	Yes
6	1985	Burkhalter and Morano [7]	21	60 days	Idiopathic	Surgical aspiration		Yes
7	1986	Borrelli et al. [34]	27	4 days		Surgical exploration and incision of corpora	1 yr	Yes
8	1988	Kimball et al. [19]	51	14 days	Idiopathic	No treatment	1 mo	Yes
9	1988	Kimball et al. [19]	37	14 days	Idiopathic	No treatment	1 mo	Yes
10	1988	Sparwasser [31]	24	35 days	Pain and jaundice due To spherocytosis	Surgical irrigation (unsuccessful)		Unknown
11	1993	De Zan et al. [33]	34	20 h	Intercourse and bent penis	Surgical exploration and drainage with necrotic tissue removed		Yes
12	1994	Ptak et al. [28]	27	2 days	Erection Former cyclist	Aspirin and propoxyphene	1 wk	Yes
13	1997	Albrecht and Stackl [5]	29	4 days	Idiopathic	Failed needle aspiration, intracavernosal etilefrine successful	1 day	Yes
14	1998	Machtens et al. [23]	44	3 days	80-mile bike ride	Heparin for 2 wk, aspirin for 6 mo	6 mo	Yes
15	1998	Thiel et al. [32]	35	1 day	Idiopathic leukocytosis	Ciprofloxin and IV heparin for 1 wk, then acetylsalicylic acid	6 mo	Yes
16	1999	Schneede et al. [30]	24	2 days	Intercourse leukocytosis	Surgical excision of transverse membrane	24 mo	Yes
17	2001	Lewis et al. [21]	24	1 day	Intercourse Sickle cell	Corporal irrigation to resolve the priapism cavernosalspongioum shunt	12 mo	Yes
18	2001	Lewis et al. [21]	33	3 days	Intercourse Marijuana, prior idiopathic priapism	Surgical exploration and Corporal irrigation	18 mo	Yes
19	2002	Pegios et al. [26]	46	10 days	Idiopathic Leukocytosis, Hepatitis A	Heparin, then acetylin silicylate	3 mo	Yes
20	2003	Goeman et al. [3]	18	3 days	A bike ride	Subcutaneous heparin for 6 wk	6 mo	Yes
21	2003	Goeman et al. [3]	27	90 days	A bike ride	Acetylsalicylic acid for 6 mo	6 mo	Yes
22	2003	Goeman et al. [3]	22	3 days	8 hour airplane flight Cyclist	Acetylsalicylic acid for 6 mo	6 mo	Yes
23	2005	Horger et al. [2]	37	2 days	Intercourse and bent penis	Pain medication and oral pseudoephedrine for 2 wk	3 mo	Yes

					Cocaine and marijuana			
24	2005	Meixel et al. [24]	21	1 day	Idiopathic	LMWH, ASA,	6 mo	Yes
25	2007	Dubois et al. [9]		3 days	Risperidone dose Protein C resistance; CRP& CK increased Ho: painful erections	low-molecular-weight heparin and ASA		
26	2008	Asbach et al. [4]	26	6h	Intercourse leukocytosis	LMWH, ASA, analgesics, ciprofloxacin	6 w	Yes
27	2008	Blaut et al. [6]	23		Idiopathic leukocytosis & increased homocysteine	LMWH, phenprocoumon	4 mo	Yes
28	2009	Kilinc et al. [18]	59		Tamsulosin dose	Surgical CS-shunt		
29	2009	Galvin et al. [10]	22	7 days	Intercourse	ASA, NSAID	3 mo	Yes
30	2010	Patel et al. [25]	21	1 day	Idiopathic	ASA, analgesics, warm compresses	3 mo	Yes
31	2011	Gluchowski et al. [11]	32	14 days	Sexual arousal	LMWH	3 mo	Yes
32	2012	Ilicki et al. [1]	20	3 days	Idiopathic Leukocytosis, incearsed LFT Excessive alcohol intake	LMWH, NSAID	6 mo	Yes
33	2012	Pepe et al. [27]	52	2 days		LMWH, ASA	2 mo	incomplete restoration of erectile function (loss of rigidity)
34	2013	Hoyerup and Azawi [14]	50		Sildenafil	Surgical evacuation		
35	2013	Hulth et al. [16]						
36	2014	Sauer et al. [29]	23	14 days	Idiopathic	LMWH, ASA		
37	2014	Hoyerup et al. [15]	70		Blood transfusion	LMWH, ASA		Danish
38	2014	Kropman and Schipper [20]				NSIAD		
39	2015	Current	26	10 hr	10 hour airplane flight	Heparin, clexane	3 mo	Yes

Table 1: Previously reported cases, age of the patients, possible causes and treatment options for PTSCC.

10 cases were surgically treated (24.4%), 21 cases (51.2%) were managed conservatively, 3 cases were treated by surgical aspiration ± injection or irrigation (7.3%), while 2 cases (4.9%) were only followed without treatment.

Nearly all patients regain painless erectile function and the mass subsided over diverse periods of time, without any remarkable complications; except one patient who failed to recover a complete erection (loss of rigidity) [27] and one patient developed difficulties in obtaining a full erection after surgical management [13]. The mean follow-up duration was 6.28 ± 6 months, and the median follow-up was 4 months.

That data show that this condition is uncommon and that the exact causes are not clearly understood. There is no clear recommendation regarding adequate management or the duration of follow-up. Therefore multicenter studies are required.

Discussion

Only 41 cases have been described including our case (Table 1). 29 cases (71%) have been described in the last two decades; this either means that there was an increase in frequency of the cases, or an increase of awareness of the clinical symptoms in combination with advanced imaging modalities, specially ultrasound and MRI. PSTCC affects mainly young men, with an average age of 32.3 ± 12.4 y. 26 (63.4%) of the patients are in the age group 20-40, rarely affecting other age groups, i. e. ≤ 20 yrs (only 2 cases/4.9%), or ≥ 60 yrs (only one case) [1,3,15].

All patients show symptoms like an acute dull aching perineal pain, a tender firm perineal mass, a rigid proximal and flaccid distal corpus cavernosum [3], almost always unilateral, and a preserved but painful erection in the majority of the patients. Within 3 days, as median of duration of symptoms, which varies from 6 hours to 3 months, the patient seeks medical advice and 91.2% of the cases less than one month. All masses are unilateral (97.6%), except for one bilateral case (2.4%) described by Lewis et al. [21].

The diagnosis is penile conditions reached by detailed history, physical examination and advanced imaging studies, but the condition should be distinguished carefully from other penile diseases e.g. penile fracture, characterized by significant penile deformity, swelling and ecchymosis "eggplant deformity" and a "rolling sign", i. e. a firm, immobile hematoma, which is palpable under the penile skin, which is rolled over; penile abscess, and penile cancer [27]. Color coded duplex sonography, MRI and CT are standard imaging studies [29]. MRI is often more useful than CT in diagnosing PSTCC and others penile conditions as MRI displays the pathologic processes in soft tissue in greater visual detail. The benefits of contrast-enhanced MRI have been demonstrated by several authors [1-3,25,27].

Recently, contrast-enhanced ultrasound has also been utilized in PSTCC diagnostics, particularly in patients with a contraindication for MRI, and it can be considered as a cost-saving alternative diagnostic tool [29]. It also helps to avoid more invasive diagnostic tools, such as cavernosography, cavernous biopsy or surgical exploration [3]. The density of the thrombus in MRI differs depending on the age of the thrombus. Firstly, in T1-weighted images, the thrombus appears hyper-intense when compared to the normal cavernous body and in T2-weighted images is hypo-intense. With formation of methemoglobin, the signal intensity gradually increases on proton density-weighted images [3].

The pathogenesis and causes of PSTCC are poorly understood. Although several risk factors have been described, the exact cause remains unclear [27]. Trauma (resulting from vigorous sexual activity, bicycling, or compression in long distance flights), hematologicals disorders (spherocytosis, sickle cell anemia, hyperhomocysteinemia, protein C resistance, blood transfusion), alcohol or drug abuse (marijuana, cocaine) and some other drugs (Tamsulosin, Sildenafil and Risperidone) have all been proposed as risk factors [9,14,18]. However, unknown causes in a large number of cases. A history of trauma, either due to sexual intercourse or cycling was present in 17 patients (41.5%).

The "two hit model" described by Ilicki et al. [1] may be explained the PSTCC pathogenesis. Firstly, transverse membrane is required, dividing the corpus into a proximal and distal portion. Hillis and Weems [13] described this membrane as a transverse fibrous septum, separating the proximal thrombotic area from the distal flaccid corpora, which is semi-permeable (due to the ability of distal part of the penis to erect), the membrane congenital or posttraumatic, unilateral or bilateral and could be identified on MRI study or during surgical exploration. Second, obstructing the permeable membrane by trigger factors (possibly micro-trauma), that may cause clotting of the blood, consequently shutting off the proximal portion. This model can justify many points e.g. the lack of occurrence of PSTCC, association with hematological disorders or micro-trauma, and may resolve spontaneously without treatment [1,13]. However, this model remains a theoretical explanation of PTSCC without histological and pathological validation.

PSTCC is not a urological crisis and several treatment options are described in the literature, ranging from conservative, or surgical aspiration ± injection or irrigation, to a follow-up observation without treatment (Table 1) [3].

Systemic anticoagulation to prevent further thrombosis and to maintain erectile function, analgesics and antibiotics are the first choice for treatment of PSTCC. LMWH and acetylsalicylic acid have been suggested for 6 months [3,32]. Various forms of surgical management, either incision of the corpora, evacuation of the thrombus, local irrigation with heparin, removal of the transverse membrane if present and cavernoso- spongiosal shunt should be reserved for cases in which conservative treatment was unsuccessful [3,21,22]. Surgical aspiration, surgical irrigation to flush out the thrombosis (unsuccessful) and fine needle aspiration followed by intracavernosal α- adrenergic agonist with satisfactory result , all considered other treatment options [5,7,31]. Close follow-up without treatment was performed in two cases with satisfactory results [19]. Our patient was treated by LMWH and acetylsalicylic acid for 6 months with rapid improvement and complete loss of pain during erection after two weeks of treatment.

PTSCC has an excellent prognosis; not only has no serious complication been reported in the literature, but also all symptoms have been resolved; the pain has gradually disappeared in all cases; the mass reduced in size and disappeared and a painless erection was recovered at diverse periods of time. Rare complications occurred, such as failure to recover a complete erection (loss of rigidity) in one case, or difficulties with a full erection after surgical management in another case [13,27].

The follow-up of the patients varies from 1 day to 24 months [5,30], with a mean duration at 6.28 ± 6 months and a median of 4 months. Follow-up of this condition should be at least 6 months during anticoagulant therapy. Our recommendation, the patient should be seen at a 3 month follow-up interval for clinical and ultrasound evaluation and at 6 months for clinical and MRI evaluation, and at 12 months for a clinical evaluation with or without MRI depend on the presence or disappearance of the mass.

Although PSTCC is not a urologic emergency and has an excellent prognosis, several points require further illustration. The considerable diversity of proposed causes and associated findings does not reflect the pathology. The relevant causes can only be identified by aggregating detailed information for each case [1]. We propose that the possible causes are collected among the following items; trauma, blood disease, alcohol or drug abuse and/or other medical drugs (Table 2).

	Number (%)
No of cases	41 (100)
1976 - 1984	5 (12.5)

1985 - 1994	7 (17.5)
1995 – 2004	10 (24.4)
2005 – 2015	19 (46.3)
Mean age, y ± SD	32 ± 12.4
Mode (range)	27 (18 - 70)
Age	
≤ 20	2 (4.9)
21 -30	26 (63.4)
31 - 40	7 (17.1)
> 40	1 (2.4)
Unknown	5 (12.2)
Causes	
Coitus or erection	11 (26.8)
cycling	6 (14.6)
hematologic disorder	5 (12.5)
alcohol or drugs abuse	3 (7.3)
long flights	2 (4.9)
Others	2 (4,9)
Unknown	11 (26.8)
Treatment	
Conservative	21 (51.2)
Surgical	10 (24.4)
injection or irrigation	3 (7.3)
No treatment	2 (4.9)
Mean follow up, months	6.4 ± 6.28 ± 6
Median, months	4

Table 2: Descriptive statistics previously reported cases of PSTCC.

Conclusion

PSTCC is a rare, non-emergency urological condition with good outcome. It affects primarily younger men who present with an acute, painful, firm perineal mass and with or without partial priapism. Color-coded duplex sonography and MRI have a crucial role in diagnosis of this condition. The pathogenesis and definitive causes are poorly understood. However, perineal trauma, blood disease, alcohol or drug abuse and/or other medical drugs may be considered as factors that can instigate this condition. Conservative management with systemic anticoagulants has an excellent outcome and surgical treatment should be restricted to cases, where conservative management fails. Systemic anticoagulant therapy should be continued for 6 months, with follow-up intervals at 3, 6 and 12 months.

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Conflict of Interest

None declared.

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