Benign transient lymphangiectasis (sclerosing lymphangitis) of the penis

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SUMMARY The literature on benign transient lymphangiectasis of the penis (BTLP) was reviewed; reports of 45 cases were collected and a further series, comprising 21 cases, is presented. Both the published and present series support the view that BTLP is associated with recent sexual activity; little support is given for an infective cause and it is confirmed that BTLP is a benign, self-limiting condition presenting in the sexually active man between 20 and 40 years old. There can be recurrences, and susceptibility to them may be encouraged by circumcision in which disturbances of lymphatic drainage may play a part. As so few cases have been reported and few were diagnosed in the present series these comments can only be pointers to further studies. The descriptions in six histological examinations, the reports as 'phlebitis' of apparently similar lesions, the report of 'sclerosing lymphangitis' in other sites, and the known histological similarity between veins and lymphatics suggest that 'sclerosing lymphangitis' is too specific a term. Until further knowledge is available, we suggest that this condition is best regarded as simple dilatation of a lymphatic vessel—namely, lymphangiectasis. As the condition is both benign and transient we propose the term benign transient lymphangiectasis of the penis for the condition formerly known as sclerosing lymphangitis.

Introduction

The presence of a painless, hard, nodular, translucent cord that suddenly appears in the penis and is usually confined to the coronal sulcus (Figure) has been known by various names (Table 1).

Of these perhaps the most usual has been 'sclerosing lymphangitis of the penis', but benign transient lymphangiectasis of the penis (BTLP) seems a better term and will be used here. BTLP is said to be rare and may present to doctors unfamiliar with it; indeed one of us (PH) first encountered it in an Emergency and Accident Department. This led to a retrospective study of BTLP and a comprehensive review of the literature. The information so collected is presented with a series of our own recent and earlier unpublished cases to stimulate further studies.

Table 1 *Nomenclature*

<i>Name</i>	<i>Author</i>	<i>Date</i>
Non-venereal lymphangitis of the penis (gonorrhoeal pseudochancres)	Hoffman	1923
Non-venereal plastic lymphangitis of the coronal sulcus	Hoffman	1938
Circular (non-venereal) lymphangitis of the coronal sulcus	Boulle <i>et al.</i>	1963
Circular indurated lymphangitis	von Berde	1937
Transient penile lymphangiectasis	Gougerot and Blum	1939
	Boyd	1970
Lymphangiectasis of the penis	Dunlop (unpublished observations)	1953
Lymphocele	Kondo and Wanatabe	1954
Sclerosing lymphangitis of the penis	King and Nicol	1975
	Shigematsu <i>et al.</i>	1965
	Lassus <i>et al.</i>	1972
	Stolz <i>et al.</i>	1974
Non-venereal sclerosing lymphangitis of the penis	Nickel and Plumb	1962
	Miki <i>et al.</i>	1966
	Rook <i>et al.</i>	1968
	Boyd	1970
	Kandil and Al-Kashlan	1970
	Greenberg and Perry	1972
	Lassus <i>et al.</i>	1972
	Janula and Janasová	1973
	Matheis	1972
		1973
	Fiumara	1975
Benign transient lymphangiectasis of the penis (BTLP)	Present series	1977

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Figure Benign transient lymphangiectasis of the penis (BTLP) in Case 8.

PUBLISHED CASES

Table 2 summarises the 45 cases recorded in the literature including those of six patients said to have lesions in non-penile sites or in whom the lesions were thought to be phlebitic. Since this report was prepared another six cases have been reported (McMillan, 1976).

The condition occurred in patients aged between 24 and 60 years (mean 32) with only one older than 50 years, and in patients in various occupations.

Marital state was recorded in only 20 cases. Nine patients were married and 11 single; the sexual partner was the wife in five cases, a regular partner in two, and a casual partner in 10. Recent prolonged or frequent coitus was admitted by 12 patients; there was no definite history of trauma, although recurrence arose in two patients after strenuous coitus.

Onset of the condition was noticed during coitus

by two patients, within 24 hours of coitus by seven patients, and within seven days by six. They presented for medical advice within hours to six weeks from onset.

Duration of the lesion was between three weeks and four months in the 37 cases in which this was recorded (mean 41 days). There were recurrences, in two patients after strenuous coitus, which lasted for 21 days in each (Greenberg and Perry, 1972; Lassus *et al.*, 1972).

Nine patients had histories of sexually transmitted disease: gonorrhoea in five, non-specific urethritis (NSU) in two, secondary syphilis and chronic prostatitis in one, and lymphogranuloma venereum in one. Concurrent infections had been present in nine patients: gonorrhoea in four and NSU in five.

Biopsy was performed in eight cases, including those of 'phlebitis', a lesion on the labium minus, and a lesion on the lip.

Various treatments were used for BTLP, including excision biopsy, aspiration, penicillin, tetracyclines, fungicides, topical corticosteroids, anti-inflammatory agents (Fiumara, 1975), wet compresses, anti-viral agents (Kandil and Al-Kashlan, 1970), and the advice to be sexually abstinent.

PRESENT CASES

Table 3 gives details of 23 episodes of BTLP diagnosed in 17 patients who attended this clinic between January 1968 and April 1975, also of four patients seen between 1951 and 1953 by one of us (EMCD). Numerical totals and means are derived from those cases in which definitive information was available. The ages of the patients varied between 18 and 51 years (mean 29) with only one patient older than 39. They had a wide variety of occupations.

Three patients were married, two were divorced, and 16 were single. The sexual partners were casual contacts, except for two cases in which contact was marital only, and four in which it was with a regular partner only. Penile trauma had occurred in two cases: a fraenal tear in one (Case 12), and repeated urethral irrigations in another (Case D).

Onset of the condition was noted by one patient during erection, by five within 24 hours of coitus, and by seven within seven days. They presented to a doctor between a few hours and 21 days after onset.

Duration (where this could be defined) varied between three and 40 days (mean 15). Recurrence other than immediately after aspiration, occurred in six patients and lasted for between seven and, in one case, more than 60 days; in the latter this was associated with an abscess of Tyson's gland.

Table 2 Summary of published reports of benign transient lymphangiectasis (or sclerosing lymphangitis) of the penis

Author	Date	No. of cases	Age range (years)	Sexually transmitted disease		Total duration (days)	Special findings
				Previous	Concurrent		
Hoffman	1923	2	Young		Gonorrhoea in both		
von Berde	1937	4	30-34		NGU-1	16-25	One contact examined, no abnormality found
Hoffman	1938	1	49	Gonorrhoea 8 years earlier	NGU	29	
Gougerot and Blum	1939	1	43	'Simple urethritis'	NSU	84	
Kondo and Wanatabe	1954	2	41-46				Aspirate contained lymphocytes in both
Braun-Falco	1955	3	24-36	Secondary syphilis-1	Chronic prostatitis—same case	21-35	Clinically similar to BTLP. Multifocal. Histology suggests phlebitis differing from Mondor's disease. Associated with tonsillitis, allergy?
Nickel and Plumb	1962	4	30-40			14-56	Biopsy in one case
Boulle <i>et al.</i>	1963	1	29			147	STS and Frei negative
							Associated ulcer. Previous topical penicillin
							Histology suggested phlebitis
Shigematsu <i>et al.</i>	1965	1	60				
Miki <i>et al.</i>	1966	3	29-49			14-30	Excision biopsy in two cases
Boyd	1970	1	24				
Kandil and Al-Kashlan	1970	2	30-35			32-42	Biopsy in one case
							Anti-viral agent tried
							Onset soon after coitus
Greenberg and Perry	1972	3	20-28	Gonorrhoea-1		21-70	One case recurred
							Onset soon after beginning coitus in one case
Lassus <i>et al.</i>	1972	4	28-38			21-56	Recurrences in one case
							Aspiration in two cases
							Biopsy in one case
Janula and Janasová	1973	1	31				
Stolz <i>et al.</i>	1974	7	24-40	Gonorrhoea-2	NSU-2	30-84	LGVCFT and Frei test positive in the case of LGV
				NSU-1	Gonorrhoea-2		Biopsy in one case
				LGV-1			Six patients professed frequent coitus
		1	29			Not known	Duration? two years in one case
						10	Lesion on upper lip
Fiumara	1975	3	17	Gonorrhoea		14-28	Lesion on labium minus. Excision biopsy
			20-28				All had coitus at least four times in preceding 24 hours. One received salicylate application

Eight patients had a history of a sexually transmitted disease: gonorrhoea in three, NSU alone in three, candidosis and NSU in one, and an abscess of a Tyson's gland in one. Concurrent conditions were present in 10 patients: gonorrhoea alone in one, secondary syphilis and gonorrhoea in one, NSU in four, and penile candidosis, genital warts, a Tyson's abscess, and urethral stricture were present in one patient each.

No biopsy was undertaken but aspirate was obtained in seven cases. Darkfield microscopical examination did not show organisms; there were few or no cells. Cultures for bacteria, Herpes simplex, *Chlamydia*, and for mycoplasmas were unsuccessful. Serological tests for syphilis gave negative results in all cases except in one of secondary syphilis and gonorrhoea (Case 17). Complement-fixation tests for lymphogranuloma venereum gave negative results in three patients as did the Frei test in one.

No specific treatment was used other than aspiration which produced temporary resolution in six

cases. Penicillin, streptomycin, and co-trimoxazole were used in one case each for coincidental infections; three patients had received local corticosteroid applications before presentation at the clinic. All patients were advised to abstain from coitus and masturbation until the condition had subsided.

PATHOLOGICAL REPORT

Biopsies have been reported in only six cases of BTLP with various findings (see Table 4). The common features were: thickening of the mural connective tissue of the lymph vessel, sometimes with fragmentation of elastic tissue (Miki *et al.*, 1966; Kandil and Al-Kashlan, 1970), partial or total obliteration of the vessel lumen possibly by organising thrombus (Miki *et al.*, 1966), and sparse evidence of chronic inflammation (Miki *et al.*, 1966; Kandil and Al-Kashlan, 1970; Matheis, 1972).

Thickening of the vessel wall with obliteration of the lumen might explain our failure to puncture the lesion in two cases, although aspiration had been

Table 3 Summary of recent cases of lymphangioectasis of the penis with earlier unpublished series

Case no.	Date of presentation	Age (years)	Circumcised	Occupation	Last coitus before onset	Duration (days)		Sexually transmitted disease		Notes
						From onset to presentation	Total	Previous	Concurrent	
1	3.1.68	36	Yes	Labourer	14 days	Not known	30	1960 Gonorrhoea		Aspirated. LGVCF†† negative
2 a	21.2.68	21	No	Engineering apprentice	1 year	2	7			
2 b	1.3.68				>1 year	Few days	>14			
3 a	9.2.70	27	No	Milkman	Few days	11	14	1969 Tyson's abscess	NSU†	Treated with co-trimoxazole
3 b	20.4.75	33			14 days	1	>60	1970 NSU†		
4 a	3.9.69	37	Yes	Unemployed	Onset during coitus	7	Not known			
4 b	29.7.71	39			Not known	3	Not known			
5 a	1.11.71	30	Yes	Labourer	7 days	<1	<3 months	1964 gonorrhoea		
5 b	4.10.72	31			10 hours	7	7			
6	4.1.72	34	No	Roadman	40 days	5	>15		Gonorrhoea	Treated with penicillin
7 a	1969	29	Yes	Student teacher	Not known	Not known	Few days			Reported by patient
7 b	9.1.72	32			14 days	Not known	>14			Aspirated
8	11.1.72	32	Yes	Cook	Few hours	2	Not known	1967 and 1971 NSU†	Candidal balanitis	Aspirated Meatal and urethral swabs for herpes and Chlamydia negative
9	4.4.72	51	No	Porter	3 days	4	20	1940 gonorrhoea		Aspirated
10	19.5.73	34	No	Inspector	Few hours	5	8	1970 NSU† and candidosis. Preputial cyst		Aspirated Attempted lymphography. LGVCF†† negative
11	25.5.73	18	Not known	Unemployed	Few hours	6	Not known			Cortisone cream
12	16.4.74	19	No	Metal worker	Not known	7		4 weeks previously torn frenum		
13	18.6.74	19	No	Seaman	Few hours	1	16			Aspirated
14 a	21.11.74	29	Yes	Fur cutter	4 days	3	3			
14 b	4.12.74				17 days	1	30			
15	22.11.74	18	Yes	Postman	14 days	21	30			Previous topical betamethasone
16	13.2.75	25	No	Postman	49 days	6	40		Small genital wart	Previous topical betamethasone
17	25.6.75	25	No	Shunter	2 days	2	Not known		Secondary syphilis. Gonorrhoea Incubating NSU†	Procaine penicillin, kanamycin
A	21.8.51	24	Not known	Labourer	2 days	1	3			
B	3.11.53	30	Not known	Docker	1 day	Not known	>15 days		NSU†	Aspirate contained protein, no cells, dark ground negative. Contact had NSC*
C	28.8.53	Not known	No	Clerk	9 days	3	16		NSU†	Given streptomycin and sulphonamide
D	1953	22	Not known	Clerk	Several months	1	Not known	1953 urethritis, 10 irrigations	Urethral stricture	Frei test negative

Table 4 Histological changes in BTLP

Characteristics	Author	Date
'Obliterating endophlebitis' with endothelial swelling, intimal fragmentation and hyperplasia, some perivascular fibrosis, some lymphocytes and histiocytes	Braun-Falco	1955
Vessel containing erythrocytes. Endothelial swelling; thickening of wall; very few inflammatory cells—said to be phlebitis	Boulle <i>et al.</i>	1963
Hypertrophy and some sclerosis of wall. Connective tissue thickening. Some inflammatory cells	Nickel and Plumb	1962
Partially thickened wall of a dilated vessel, containing amorphous eosinophilic material. No inflammatory cells	Miki <i>et al.</i>	1966
In a second case, the above findings with scanty elastic tissue, some fibrosis and possible organising thrombus	Kandil and Al-Kashlan	1970
Marked thickening, almost with total occlusion and loss of differentiation between intima and lumen. Some local inflammatory infiltrate		
Marked thickening of wall with oedema of connective tissue. A very few inflammatory cells	Lassus <i>et al.</i>	1972
Penis. An unusually hypercellular oedematous fibrous reaction in mural connective tissue, fragmented elastic lamina and some perivascular lymphocytes and histiocytes	Stolz <i>et al.</i>	1974
Labium minus. Very thickened wall, obliteration of the lumen, sporadic mononuclear cell infiltrate		

successful in several of the published cases, and in seven of the present cases; aspiration resulted in disappearance of the lesion, albeit usually with rapid recurrence.

Of the generally recognised disorders of lymphatic vessels, the histological changes of 'sclerosing lymphangitis', and 'thrombolympfangitis productiva' (Pfleger *et al.*, 1967) most closely resemble those of BTLP. However, the disappearance of the vessel on aspiration followed by rapid recurrence, the characteristic translucency, and the lack of chronic inflammatory changes suggest simple dilatation of the lymph vessels. In Most's classification of lymphatic swellings (Kondo and Wanatabe, 1954) this would be 'lymphangiectasis'—a term already used for describing the characteristic clinical entity (Dunlop, 1953, unpublished observations; Kondo and Wanatabe, 1954). This term may be more correct than sclerosing lymphangitis; or perhaps there may be two processes with sclerosis developing after a period of intermittent, recurrent, or persistent lymphangiectasis. It may be relevant that biopsy, two weeks after onset (Miki *et al.*, 1966), showed fibrosis as a more notable feature, than did biopsy only a few days after onset (Kandil and Al-Kashlan, 1970).

Nickel and Plumb (1962) stated that their histological data 'demonstrated for the first time that

the lesion is a result of a fibrous thickening of a large lymphatic vessel'. However, venous and lymphatic vessels can appear indistinguishable (Boulle *et al.*, 1963; Lever, 1967). Boulle *et al.* (1963) cite their own case of apparent BTLP in which histology and the presence of intraluminal erythrocytes suggested 'phlebitis' rather than 'lymphangitis'. Braun-Falco (1955) described a clinical appearance identical with that of BTLP in which superficial cord-like thickenings were present in the penis, yielding serous fluid, and in which the biopsy appearance suggested 'obliterating endophlebitis'. Boulle *et al.* (1963) thought that this clinically resembled Mondor's phlebitis (Mondor and Bertrand, 1951), although it differed histologically; they suggested that hypersensitivity to infective agents was a possible cause. The penile lesions in these reports of phlebitis very closely resemble those of BTLP. Perhaps the clinical appearance of BTLP may be caused by venous or lymphatic lesions, or both. Lesions similar to BTLP have been reported in the upper lip of a man and in the labium minus of a woman, and Stolz *et al.* (1974) considered that infection was the most likely reason for these changes.

Discussion

CAUSATION

Most authors have accepted that BTLP is not caused by infection, ascribing particular importance to trauma during recent vigorous sexual activity, although they speculate about possible infective causes.

TRAUMA

Postulated traumatic causes include mechanical irritation that allows entry of pathogens into the mucosa, high coronal pressures during the plateau phase of sexual excitement (Matheis, 1972), traumatic obstruction of the vessels (Lassus *et al.*, 1972), activation of resident viruses (Kandil and Al-Kashlan, 1970), and irritation by menstrual blood.

Possible undue trauma occurred in only two cases: one followed 10 irrigations for persistent urethritis (Case D, Table 3), and one followed one month after a fraenal tear (Case 12, Table 3). All patients had had coitus, and in many the onset of BTLP occurred a few days later. Particularly in the reports by Greenberg and Perry (1972), Lassus *et al.* (1972), Stolz *et al.* (1974), and Fiumara (1975), patients professed recent and frequent intercourse—up to six times a day. This was a less striking feature in our patients of whom 12 had had coitus within the week before onset of BTLP.

If trauma during coitus or masturbation is responsible, then BTLP should surely be more common than it is. Naturally, it is particularly those patients who notice penile lesions after sexual activity who present to venereologists, and we wonder if the condition presents in other ways.

INFECTION

Suggested infective causes include viruses either 'resident' or freshly acquired (Kandil and Al-Kashlan, 1970), susceptibility after previous or concurrent sexually transmitted infections, and agents—such as members of the *Chlamydia* genus and mycobacteria.

The finding of a similar clinical lesion in the upper lip of a man after orogenital contact, and a lesion on the labium minus in which the histological findings resembled those of BTLP, suggest that the lesion may occur at non-penile sites because of infection (Stolz *et al.*, 1974). However, the histological similarity between veins and lymphatics (Lever, 1967), the existence of Mondor's phlebitis, and the questions raised by Boule *et al.* (1963) discussing the work of Braun-Falco (1955), suggest that the existence of multifocal BTLP with its implications of transmission has yet to be proved.

Previous episodes of sexually transmitted infection had occurred in nine of the published cases with concurrent infections diagnosed in nine cases; in our series seven patients (eight presentations) had had previous infections, and eight (nine presentations) had concurrent sexually transmitted infections. Such conditions would be expected in these patients, but no close relationship is apparent between the onset or recurrence of BTLP and previous or concurrent sexually transmitted diseases. Little information is available about sexual contacts in the published reports; examination of the sexual partners of our patients yielded no helpful information.

The failure to culture any infective agent from the aspirate obtained from our patients is perhaps not surprising in view of the few cells in the fluid; our failure to culture herpes simplex virus militates against this as a likely candidate for a resident virus activated or reactivated by trauma during sexual activity.

CIRCUMCISION

Trauma seems to play a part in some cases and perhaps a local anatomical abnormality is concerned. The normal anatomy of penile lymphatics (Lockhart *et al.*, 1965) has not been studied by us but we made one unsuccessful attempt to inject contrast medium for radiography into the lumen of a lesion. Lymphography has been used in studies of other penile

conditions (Jantet, 1962; Janca, 1972) and study of BTLP by this method is indicated.

It is remarkable that in no published report is circumcision specifically mentioned for it is possible that circumcision may disturb the normal lymphatic drainage of the glans and prepuce, making the trauma of coitus more likely to provoke BTLP. All of our patients except one were Caucasian, and in a random sample of our clinic attenders we would expect 25% to be circumcised (Taylor and Rodin, 1975). Information on this was recorded for 17 patients and seven (41.2%) were circumcised. Four of these seven patients suffered recurrences, as did two of the uncircumcised patients.

INCIDENCE

The seeming rarity of this condition has been stressed (Rook *et al.*, 1968; Greenberg and Perry, 1972; Matheis, 1972). Certainly BTLP is likely to be missed by patients and doctors; it is often diagnosed incorrectly or not observed because of its painless, self-limiting nature. Reports include: four cases seen in six years (Nickel and Plumb, 1962), four cases seen in 18 months (Lassus *et al.*, 1972), and three cases seen out of 10 000 attenders (Fiumara, 1975). In our series 15 patients presented with this condition (of which four were recurrences) between January 1972 and April 1975, during which time about 22 000 diagnoses were made for men. Our cases seemed to occur in clusters, presumably because of improved recognition of BTLP by the clinic staff once a case had been demonstrated.

All patients in the present series had experienced coitus; they were between 18 and 60 years of age (mean 29); there are three cases reported in the literature and one in the present series in which the patient was older than 40. All our patients were heterosexual, all except one were Caucasian; this was in a clinic where 43% of men are non-Caucasian. Patients had a wide variety of occupations both in the published and present series. Initial presentation was usually to a venereologist, but six of our patients presented first to their family doctors and two to an Emergency and Accident Department.

COURSE

The lesion usually occurred within seven days of coitus; the patients presented for medical advice at any time from a few hours to weeks after the onset. Total duration, where this could be defined, was from three days to five months (mean 41 days in reports, and 15 days in our cases), but one case possibly lasting two years has been reported (Stolz *et al.*, 1974).

Spontaneous resolution will occur in all cases;

cure by excision biopsy is immediate, while aspiration produces resolution, usually temporarily. Various anti-bacterial, anti-inflammatory and corticosteroid preparations, as well as wet compresses, silver nitrate and diathermy, have been tried without beneficial effect.

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