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NON-VENEREAL SCLEROSING LYMPHANGITIS OF THE PENIS

A Clinicopathologic Treatise

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Abstract. Two cases of non-venereal sclerosing lymphangitis of the penis (NSLP) are reported. The response of the 2 reported cases to N^1, N^1 anhydrobis-(B-hydroxyethyl) biguanide hydrochloride (ABOB), an antiviral agent, may possibly denote a viral aetiology of NSLP. Histologic examination of 1 of the reported cases conforms with the only available report of Nickel & Plumb.

Non-venereal sclerosing lymphangitis of the penis (NSLP) first described by Hoffman in 1923 (1), is a very rare condition; only 12 cases have yet been reported (3) and although symptomless, usually causes the patients much anxiety. The paucity of NSLP and the interesting rapid initial response of the 2 reported cases to the antiviral agent N^1, N^1 anhydrobis-(B-hydroxyethyl) biguanide hydrochloride (ABOB) (Flumidin, AB Kabi, Stockholm, Sweden) has prompted the publication of this paper. Moreover, it was thought worthwhile to report the histology of NSLP as seen in one of our patients in view of the fact that a histologic description of NSLP was reported only once by Nickel & Plumb (3).

CASE REPORTS

Case 1. A 30-year-old married man, working as a male nurse in a medical ward, manifested a symptomless cord-like lesion of the penis which he noted some hours after sexual intercourse though it was not possible for him to ascertain inception precisely. However, the patient was sure that 4 days before consultation it was entirely absent. On examination, a cord-like subcutaneous lesion could be seen and felt encircling almost three-quarters of the penile shaft just behind the corona of the glans penis and of about 2 mm thickness (Fig. 1). The skin over the lesion was normal in every respect. The lesion itself was not tender and was not fixed to the skin or to the deeper structures and no edema was noted. Enquiry of the patient's sexual habits revealed no abnormality except that he contacts his wife almost daily. The patient was subjected to routine investigations, including blood kahn, but nothing relevant was noted. The rare possibility of calcified medina worm affection of the genital region (5) prompted X-ray examination of the penis but no calcified shadow was seen. The patient was referred to the surgeon for excision biopsy; the wound gaped after removal of the stitches and a new, 1½ cm long lesion was noted.

Case 2. A 35-year-old man manifested a rather convoluted symptomless lesion of the penis of 2 days duration which he noted after sexual intercourse. On examination, a worm-like lesion could be seen only on stretching the skin of the penis just behind the corona but could be easily felt as a fleshy, worm-like, not tender, freely mobile subcutaneous lesion about 3 mm thick. There was slight edema of the organ around the lesion but no overlying erythema.

HISTOLOGY

Histologic differentiation between veins and lymphatics may be difficult unless some erythrocytes are seen. The examined material, apart from the clinical impression, was justifiably considered a lymphatic in view of the difference of arrangement of elastic fibers from those usually seen in veins which run concentrically in the media.

The cut section of the vessel showed marked thickening with almost complete occlusion of the vessel lumen (Fig. 2). Special staining for elastic tissue (Weigert) showed loss of differentiation between the intimal layer and the connective tissue within the vessel lumen thus appearing as a homogenous material devoid of nuclei being unstainable with Weigert method (Fig. 3). A third section, cut obliquely at the level of a valve, demonstrates the connective tissue occluding the lumen with moderate numbers of histiocytes, fibroblasts and lymphocytes, together with invading blood capillaries simulating a thrombus in the process of organisation (Figs. 4 and 5). The inflammatory cellular infiltrate in and around the lymphatic vessel is not sufficiently dense to accept the process unequivocally as a non-specific inflammatory one.

Fig. 1. (case 1). Cord like lesion.

TREATMENT

Case 1 was advised to take 800 mg (two tablets) of ABOB ter die sumendum, Case 2 was advised to take 800 mg bis in die for a week but later was advised to increase the dose to 800 mg ter die sumendum.

Case 1 was completely cured within 30 days. Case 2, under the initial ABOB low dose, showed rapid regression of the edema and the lesion itself acquired the typical semi-cartilaginous hardness. Its thickness was also reduced to about 2 mm. Complete cure was, however, achieved within 40 days.

DISCUSSION

On clinical grounds, it was felt from the start that we were not dealing with a vein pathology, since, anatomically, no subcutaneous vein runs circumferentially at the site involved (2). Furthermore, thrombophlebitis could be excluded because of the absence of clinical symptoms and

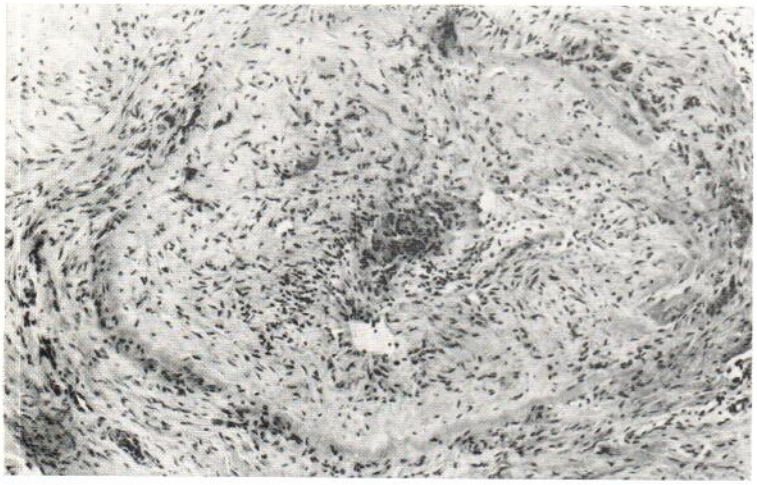

Fig. 2. Occluded lymphatic vessel with a slight to moderate cellular infiltrate. Hematoxylin-eosin, × 40.

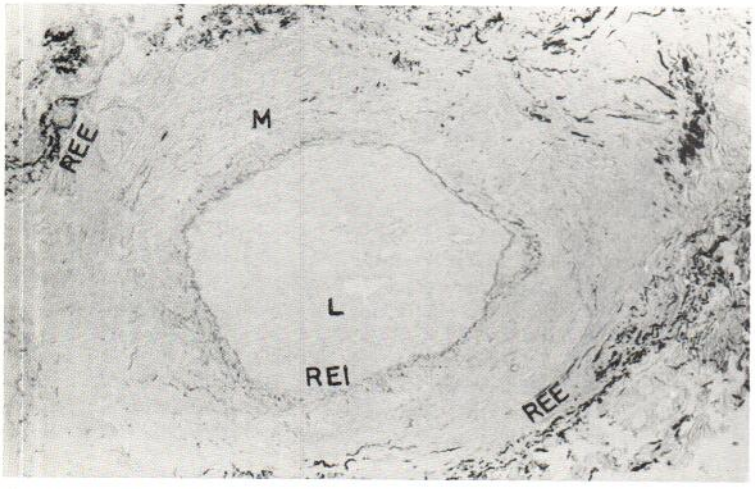

Fig. 3. Elastic stain. *L*, lumen; *REI*, rete elasticum internum; *REE*, rete elasticum externum; *M*, muscle layer. Weigert stain, × 80.

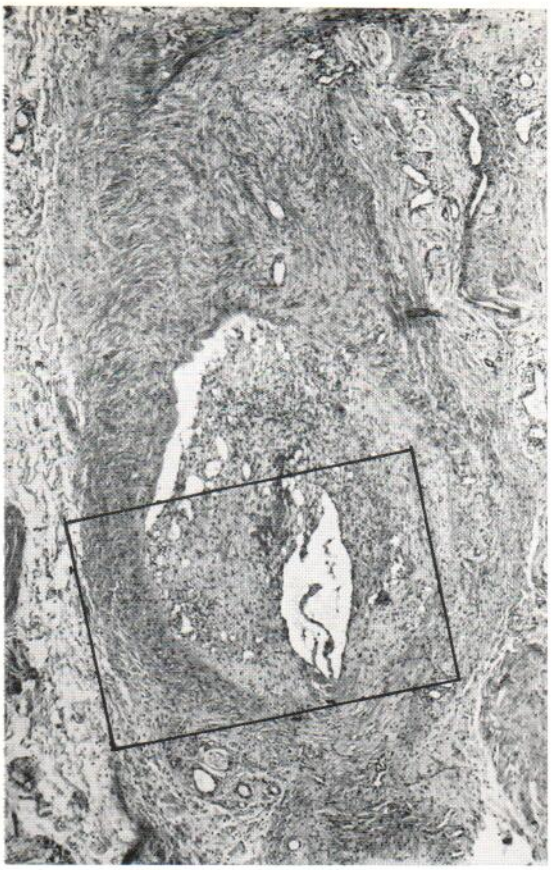

Fig. 4. Oblique section of the lymphatic vessel. HE, × 46.

signs of inflammation. The clinical evolution of case 2 is noteworthy and indicates the existence of a symptomless edematous presclerotic stage in which the lesion was rather fleshy and with slight, adjacent edema which eventually subsided. The lesion then became sclerotic, as is usually described.

The aetiology of NSLP is as yet unknown but it was suggested that it may be related to mechanical trauma, virus infection, irritation of menstrual blood or tuberculosis (3). Although Nickel & Plumb did not approve any of the aforementioned aetiologic factors, we feel that NSLP may be attributed to a virus infection triggered by sexual intercourse or minor trauma. This suggestion is prompted by the relative prevalence of NSLP in medical staff (of the 14 cases hitherto reported, including the 2 present cases, 4 were medical staff) and by the interesting sequence of events of case 1: excision, development of a new lesion of NSLP after the trauma of the surgery and its rapid involution under ABOB therapy, possibly denoting a virus infection precipitated by trauma. Although NSLP is a self-limited disease, it is felt that the response of the two cases may be considered specific because of the rapid initial response to ABOB. It is clear that this suggestion has to be confirmed before being considered true. The true relationship between sexual intercourse and NSLP remains obscure and the usual history of NSLP being noted after sexual intercourse may be relevant as a form of minor trauma, or may be a mere coincidence being only noted because of habitual washing after coitus.

The histologic features of NSLP, as shown in the presented case, simulate what Pfleger et al. have termed "Thrombolymphangitis productiva" in their histologic study of 900 cases of lympha-

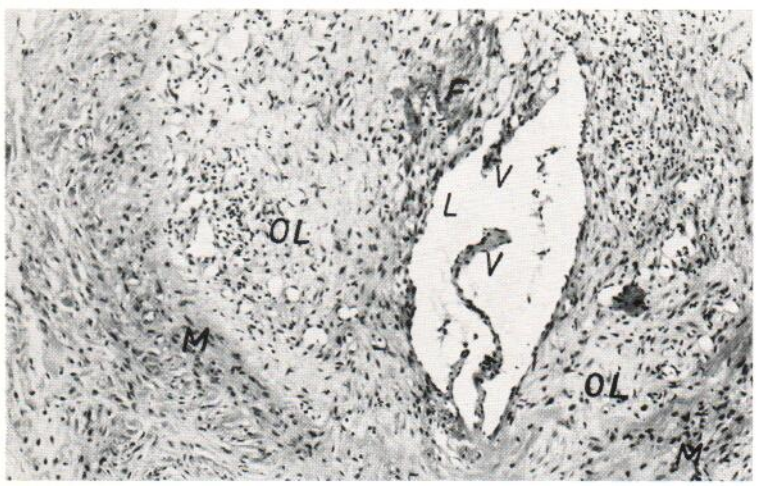

Fig. 5. Enlargement of Fig. 4. *V*, valve; *F*, fibrin; *M*, muscle; *L*, lumen occluded by connective tissue; *OL*, occluded lumen in process of organisation showing numerous histiocytes, fibroblasts, lymphocytes and invading blood capillaries. HE, × 120.

tics of the lower limbs (4), possibly indicating that no inflammatory process is implicated; a thrombotic process is suggested.

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