Virchows Arch. A Path. Anat. and Histol. 377, 79–84 (1977)

Virchows Archiv A

Case Report

Congenital Testicular Lymphangiectasis

Manuel Nistal* and Ricardo Paniagua**

Department of Morphology, School of Medicine, Autonomous University of Madrid, and Department of Pathology, "La Paz" Hospital, Madrid, Spain

Summary. Testicular lymphangiectasis are described for the first time in a patient with bilateral inguinal cryptorchidism. A great number of irregular lymphatic channels was observed within the parenchyma and the tunica vasculosa in both testes. Large and numerous anastomosis between the lymphatic vessels of these two areas could also be seen. The MTD and the TFI of the left testis were normal. Both parameters were very low in the right testis. The association of this fact with the greater development of the lymphatic vessels in this testis strongly supports the idea that testicular lymphangiectasis interfere mechanically with the testis tubular development.

Key words: Lymphangiectasis – Testis – Cryptorchidism – Lymphatic vessels.

Introduction

Congenital lymphangiectasis bear different clinical-pathological features. In the case of generalized abnormal development of the lymphatic vessels, the patients can show body hemihypertrophy, malabsorption and respiratory failure (Noonan et al., 1970). However, the involvement of a single organ is more usual. In these cases, some clinical findings become characteristic: There is a loss of albumin accompanying intestinal lymphangiectasis (Waldmann et al., 1961), and a respiratory distress in the case of congenital pulmonary lymphangiectasis (France and Brown, 1971; Nistal et al., 1973).

We describe here, for the first time to our knowledge, testicular lymphangiectasis in a child with bilateral cryptorchidism.

* Profesor Agregado de Histología y Embriología
** Profesor Adjunto de Histología y Embriología

For offprints contact: Dr. Manuel Nistal, Departamento de Morfología, Facultad de Medicina, Universidad Autónoma, C/Arzobispo Morcillo, 4, Madrid-34, Spain

Case Report

A 7 years old child was admitted to the hospital to be treated for cryptorchidism. He showed a normal psychomotor development. There was no history of respiratory failure or malabsorption. All the clinical findings were normal except for the abnormalities to be described in the genital tract. Thorax X-ray and laboratory test were normal. The sexual chromatin was negative and the cariotype was 46, XY. The basal testosterone level was 2.25 μg/ml and went up to 9.60 μg/ml after HCG stimulation.

Both testes could be palpated at the inguinal duct. Their size, measured during the operation, was found to be a little diminshed. There were no abnormalities at the epididymis and at the spermatic cord.

Material and Methods

An orchidopexy was performed and biopsy samples were taken from both testes. The testicular samples were fixed in buffered 10% formaldehyde during 48 h and embedded in paraffin. Sections of 6 μm thickness were obtained and stained with H.E., PAS and Masson trichrome. Both, mean tubular diameter (MTD) and Mack's fertility index (TFI), were determined in both testes.

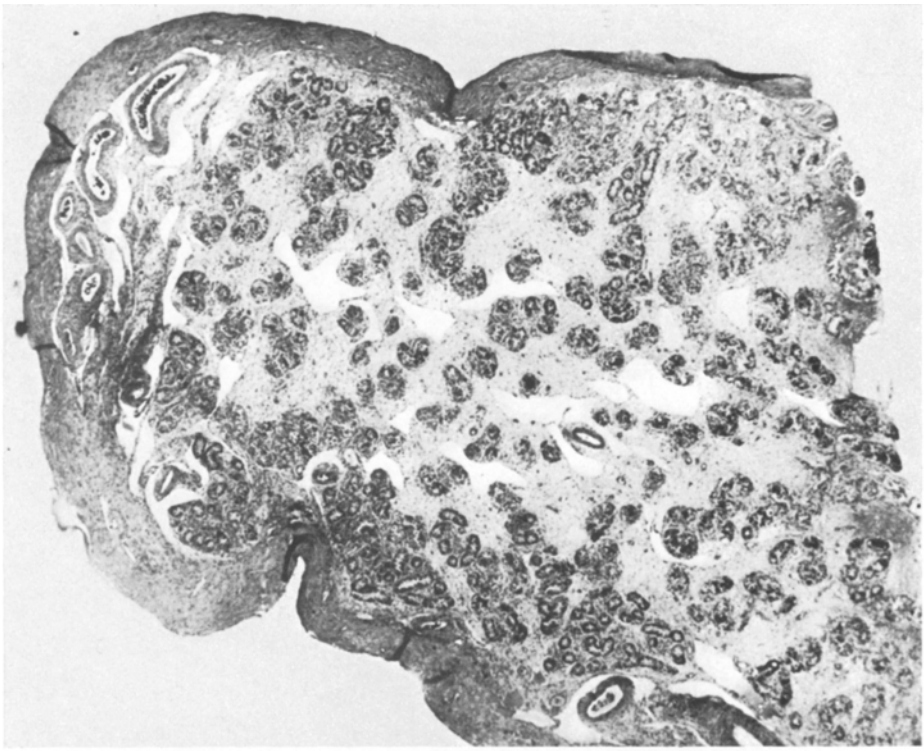
Results

Right Testis

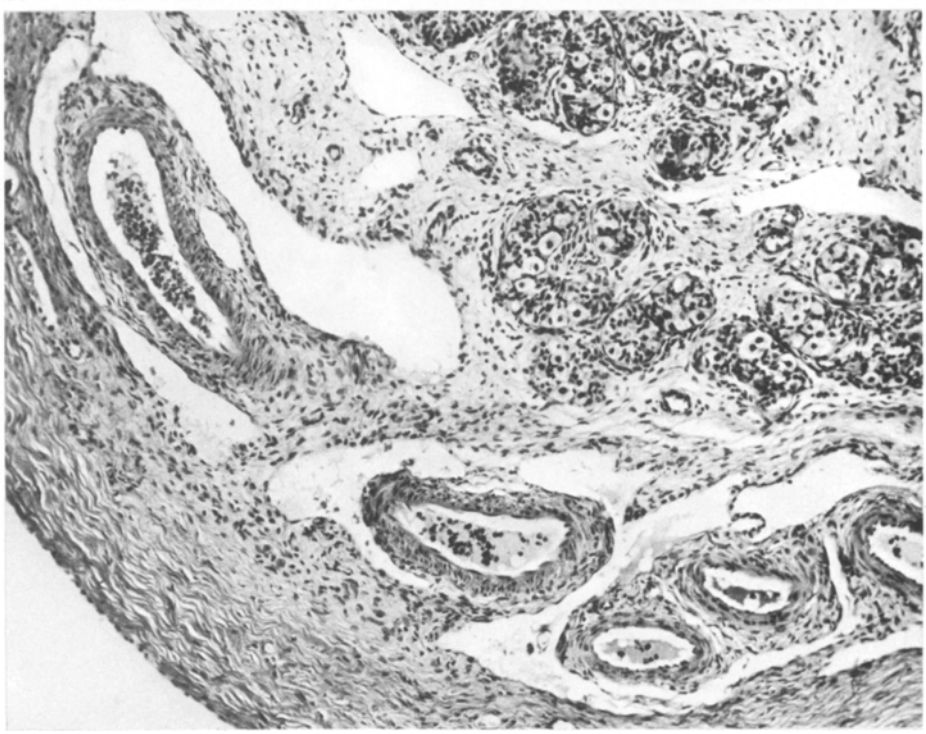
The thickness of the testicular capsule showed variations in connection with the alterations of the tunica vasculosa. The latter is made up by loose connective tissue including arteries, veins and lymphatic vessels. The lymphatic vessels gained greatly in size. They surrounded the arteries and veins in such a way that these blood vessels kept in connection with the connective tissue only by very fine pedicules (Figs. 1, 2). The MTD was only 50 μm and the TFI was 62%. The basal membrane was not thickned. The interstitial tissue was increased, partly by the presence of loose connective tissue, and partly by the development of lymphatic vessels (Fig. 3). These vessels were seen as irregular channels, lined by flattened cells, and containing a slightly eosinophilic material in their lumen (Fig. 4A). The testicular parenchyma was distorted by the presence of these lymphatic vessels. Extensive comunications between parenchymal and subcapsular lymphatic vessels were seen on serial sections.

Left Testis

The tubular and interstitial alterations were smaller than those observed in the right testis. Macroscopically, their size was nearly normal. The MTD was 62 μm and the TFI 80%. Dilated lymphatic channels were irregularly distributed within the parenchyma. Similar lymphatic vessels were seen in the tunica vasculosa in a smaller number than those observed in the right testis (Fig. 4B).



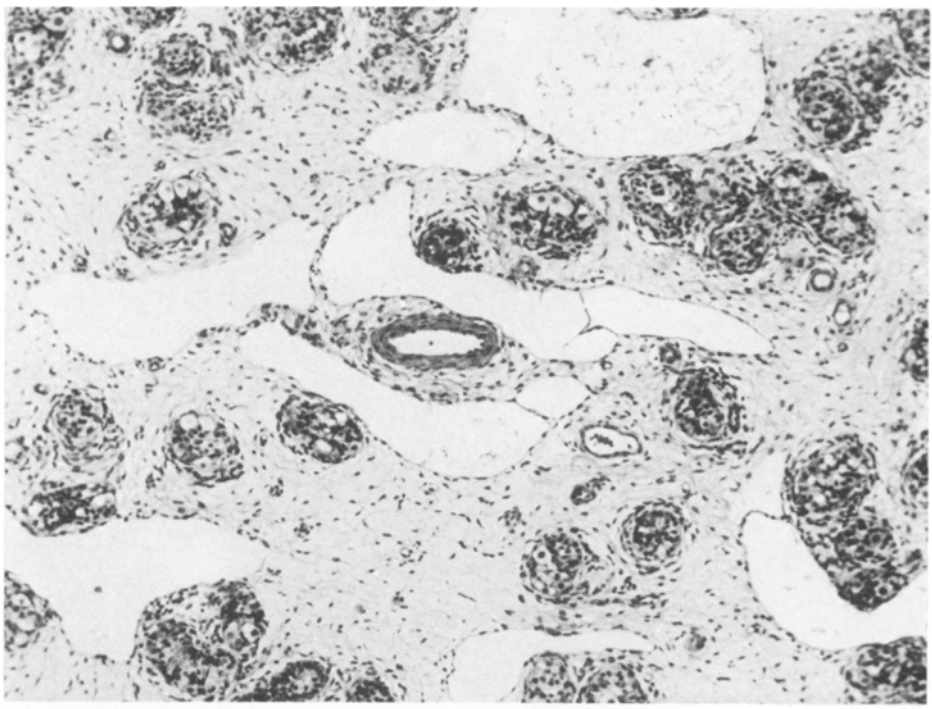
1



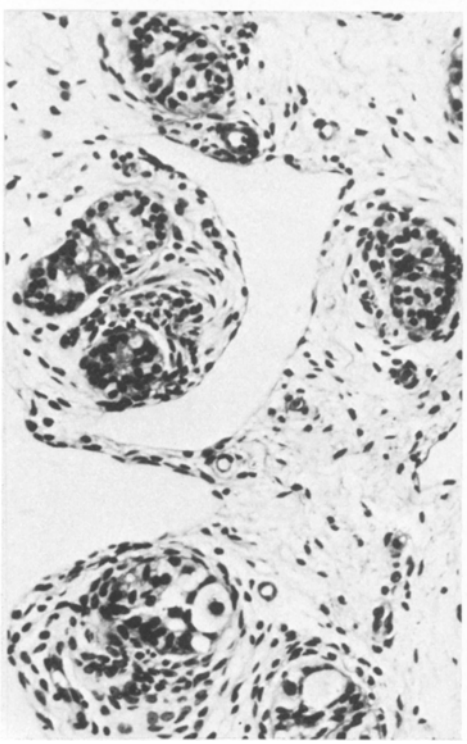
2

Fig. 1. Whole section of left testis biopsy. The development of the seminiferous tubules is scant. Dilated lymphatic vessels are seen in the lobuli testis as well as in the subcapsular area. H.E. $\times 30$

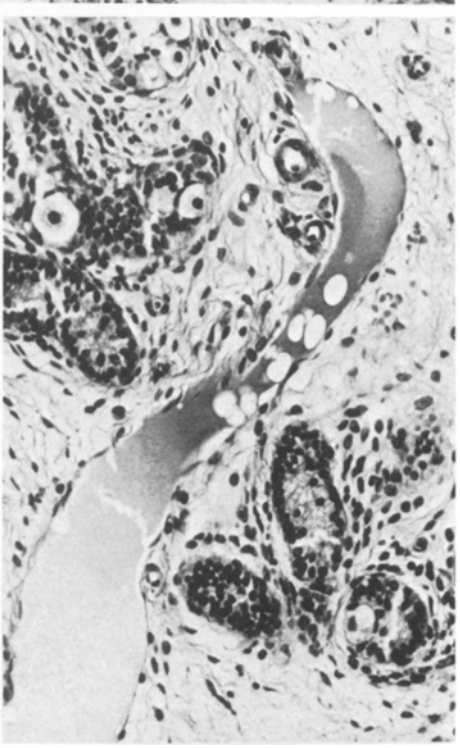
Fig. 2. Abnormal development of lymphatic vessels in the tunica vasculosa. Several arteries and veins appear surrounded by lymphatic channels. H.E. $\times 250$



3



4A



4B

Fig. 3. The interstitial tissue is wide. It contains dilated or cystic lymphatic vessels which distort the lobular architecture. H.E. $\times 250$

Fig. 4. A Seminiferous tubules partially surrounded by lymphatic channels. The tubular diameter (MTD) and fertility index (TFI) are diminished. H.E. $\times 500$. **B** Markedly dilated lymphatic vessel containing granular eosinophilic material. H.E. $\times 500$

Discussion

The development of the human testicular lymphatic system begins early in the foetus. Ostroverkhova (1860) showed the presence of isolated lymphatic capillaries within the testis of several foetus. Isolated capillaries in the lobuli testis and a lymphatic network in the septula testis were observed during infancy. The development of peritubular lymphatic vessels become complete just before the onset of puberty. In the human adult testis, the lymphatic capillaries make up a peritubular network that drains through collector vessels which run through the septula testis along the blood vessels. These collector reach either the albuginea or the rete testis (Hundeiker, 1969).

The resticular lymphangiectasis is a congenital anomaly due to an abnormal development of the lymphatic vessels of the testis. This hypothesis is supported by the fact that there is no accompanying pathology which indicates the existence of an obstruction to the lymphatic flow at the level of the spermatic cord or in the regional lymph nodes.

The fact that the testicular lymphangiectasis are not found together with generalized lymphangiectasis allow one to consider them as a separate entity.

The relationship between testicular lymphangiectasis and bilateral cryptorchidism described in this case is an occasional finding. This is supported by the fact that in large series of histological studies on undescended testes, lymphangiectasis were not observed (Scorer and Farrington, 1971).

The influence of testicular lymphangiectasis upon the testis development becomes difficult to evaluate because of the associated cryptorchidism. It is common to find small testes in inguinal bilateral cryptorchidism with low MTD and TFI, but the extent to which both testes are affected is similar. The MTD and TFI of the left testis of this patient were practically normal, contrasting with the marked diminution of both parameters in the right testis.

As the abnormal development of the lymphatic vessels is more apparent in this testis, it would be possible that the lymphatic obstruction had somehow, probably mechanically, interfered with the tubular development.

The prognosis is not good as far as fertility is concerned. It is bad in bilateral cryptorchidism and even worse if surgical correction is done late. Nevertheless, in the present case, and considering that the size of the testis, the MTD and the TFI are not excessively low, one might expect an acceptable spermatogenesis; but the difficulty of the tubular development due to the presence of lymphangiectasis makes the prognosis worse.

References

France, N.E., Brown, R.J.K.: Congenital pulmonary lymphangiectasis. Arch. Dis. **46**, 528–534 (1971)

Hundeiker, M.: Lymphatic vessels in the parenchyma of human testis. Arch. Klin. Exp. Derm. **235**, 271–276 (1969)

Nistal, M., Belaustegui, A., Quero, J., García-Sánchez, Pérez-Martínez: Las linfangiectasias pulmonares congénitas como causa infrecuente de distress letal en el recién nacido. Med. Clin. **61**, 616–623 (1973)

Noonan, J.A., Walters, L.R., Reves, J.T.: Congenital pulmonary lymphangiectasis. Amer. J. Dis. Child. **120**, 314–319 (1970)

Ostroverkhova, V.G.: Macro-microscopicheskoe issledovanie vnutriorgannoi limfaticheskoi sistemy muzhskoi polovoi zhelezy (Macromicroscopic study of intra-organ lymphatic system of male gonad in man). Arch. Anat. Gist. Embriol. **39**, 59–65 (1960)

Scorer, C.G., Farrington, G.H.: Histological studies of the undescended testis. In: Congenital deformities of the testis and epididymis, pp. 58–75. London: Butterworth and Co. 1971

Waldmann, T.A., Steinfeld, J.L., Duchter, J.F., Davidson, J.D., Gordon, R.S.: The role of the gastrointestinal system in "idiopatic hypoproteinemia". Gastroenterol. **41**, 197–207 (1961)

Received May 21, 1977