Case Report

Neonatal Testicular Hemangiolymphangioma: A Case Report

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Abstract

A 30-day-old neonate was brought to our hospital due to testicular neoplasm in the right scrotum. Scrotal ultrasonography revealed a mixed cystic and solid mass in the testis. Analysis of testicular tumor markers was negative. Scrotal exploration was performed. A red nodular tumor was removed from the testis by surgery. Histological examination of the specimen showed it to be hemangiolymphangioma (HLA).

Keywords: Hemangiolymphangioma (HLA), neonate, testis, ultrasound

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Introduction

Neonatal testicular tumors are rarely encountered in clinic. Hemangiolymphangioma (HLA) is a mixed vascular malformation (VM) with both vascular and lymphatic elements,[1] usually caused by lymphatic-venous or lymphatic-capillary malformation. It is difficult to diagnose HLA except through histological examination. It is benign in most cases.[2] Here, we report a case of neonatal testicular HLA in a 30-day-old infant.

Case report

General information

A 30-day-old neonate was admitted to our hospital. The mother's pregnancy was normal and the baby was delivered in a local hospital by cesarean section without any complication. At birth, the baby weighed 4,500 g and was healthy except for a mass in the right scrotum. He was the third baby in his family. There was no history of genital abnormality in his siblings. The ultrasound report from the local hospital suggested that the mass in the right scrotum was highly suspicious for testicular tumor, and a hydrocele testis was observed in the left scrotum. The baby's parents approved the report of this case.

Clinical examination

The baby was examined by an urologist. The skin temperature was normal and there was no wound on the skin of the testis. The right side of the scrotum was violet-black in color. The right testis itself was not involved, but a neoplasm about 4 cm × 4 cm × 4 cm in size was felt in the scrotum, which did not appear to have changed since birth. The neoplasm was firm in texture and painless to pressure. The cystic neoplasm in the left scrotum also did not cause pain when touched. The test of perviousness to light was positive. The location and size of the left testis in the scrotum were normal. The urine parameters were within normal range.

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The scrotum was further examined with a Toshiba Aplio500 scanner and PLT-805AT (8MHz) high-resolution linear-array transducer. A longitudinal color Doppler ultrasonogram of the left scrotum showed that the left testis was normal and homogeneous with an obvious and regular flow signal, while a clearly defined testicular image was not observed in the right scrotum. The left testis was measured at 1.7 cm × 1.4 cm and the right neoplasm was 3.5 cm × 3.6 cm × 3.5 cm. The hydrocele was about 1.4 cm deep surrounding the left testis (Figure 1A). The boundary of the neoplasm was fuzzy and indistinct. The echo in the neoplasm was mixed cystic and solid. The echo of the solid part was medium, while the liquid portion partially contained tiny dots. A small cystic area and calcification were also observed within the neoplasm (Figure 1B). No flow signal was found (Figure 1C).

The transverse color Doppler ultrasonogram of the right scrotum showed the testis and the right neoplasm (Figure 1D). The size of the lymph node was within the normal range, with a clear border and low internal echo in the bilateral inguinal region.

Surgery

The baby was anesthetized with Sevoflurane and laid on the bed. After disinfection, a 3-cm diagonal incision was made by the surgeon on top of the scrotal skin. The spermatic cord was found, with a diameter of 0.5 cm. The testicular sheath membrane capsule was opened, and the mass was about 3.0 cm × 3.0 cm in size. The external tunica vaginalis was complete. No testicular parenchyma was found. The spermatic cord and its attachments were separated and the spermatic cord was ligated at high position. The entire mass and part of the spermatic cord were removed after pathology reported a benign mass.

The size of the lymph node in the bilateral inguinal region was normal. Necrotic tissue was observed inside the mass and some brown liquid squirted out when it was cut open (Figure 2A). The mass was cystic with many cavities (Figure 2B). Hematoxylin and eosin staining showed that the capsule wall and cysts were lined with simple squamous epithelium. Necrosis was observed in the contorted seminiferous tubule, and hemorrhage and calcification were also found (Figures 2C and 2D). Thus, pathology diagnosed testicular HLA with necrosis and calcification.

The patient recovered quickly. At one year of follow-up, no recurrence of the mass was observed.

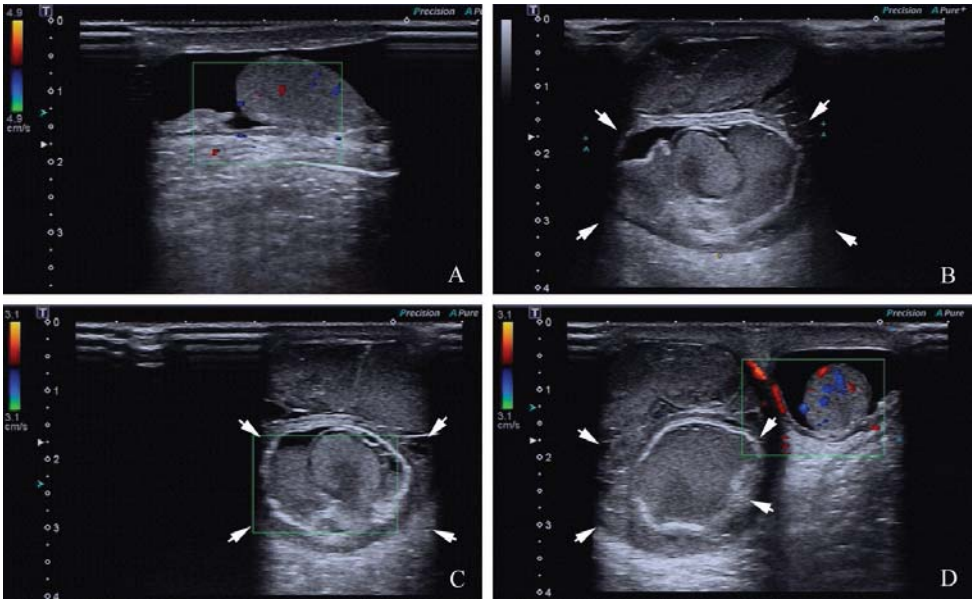

Figure 1. Longitudinal color Doppler ultrasonograms. **(A)** Left testis was normal and homogeneous with a hydrocele and regular flow signal. **(B)** The neoplasm could be observed in the right scrotum. The boundary of the neoplasm was indistinct (indicated by arrows). Echo of the neoplasm was mixed cystic and solid. A medium echo was detected from the solid part, while the liquidity portion was partly in tiny dots. A small cystic area and calcification were observed within the neoplasm. **(C)** The right scrotum showed no flow signal. **(D)** Left testis and right neoplasm.

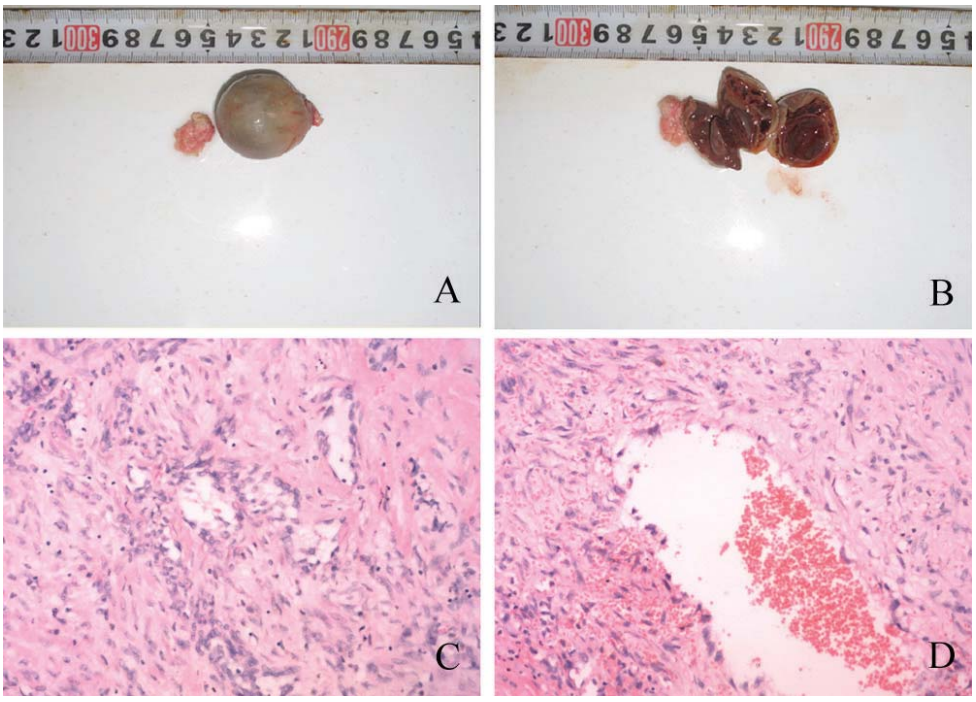

Figure 2. The testicular neoplasm. **(A)** The diameter of the resected neoplasm was 3.2 cm. **(B)** The mass was cut open, we see the internal structure of the mass. **(C)** Hematoxylin-eosin staining. (H&E,×100). **(D)** Hematoxylin-eosin staining (H&E,×100).

Discussion

The majority of testicular tumors are malignant. Seminoma is a predominant tumor type in adults and yolk sac tumor is dominant in children.[3] Benign testis tumors include approximately 5% of all cases of testicular neoplasms, including lipoma, interstitial cell tumors, hemangioma and adenomatoid tumors. HLA is much rarer than others.[2]

HLA is an extremely rare malformation of the lymphatic system and blood vessels[1] in which the vascular system consists of blood vessels and lymphatic vessels. Based on clinical, histological, and cytological features, Mulliken and Glowacki[4] differentiated vascular anomalies as either tumors (including infantile hemangiomas) or other malformations (vascular or lymphatic). Clinically, most vascular anomalies are VM. Hemangiomas are characterized by endothelial hyperplasia with rapid post-natal growth and slow involution, while flat endothelium characterizes VM.[5] Mulliken's and Glowacki's[4] classification has been applied

to lymphatic anomalies. Lymphatic malformations have a single layer of endothelial cells, and the lymphangioma is the post-natal proliferative phase.[5]

HLAs are mostly benign and occur in a variety of anatomic locations, including oral, maxillofacial, axilla, abdominal cavity and extremities, but rarely in the spermatic cord or scrotum.[6] Although HLA can be diagnosed via histology of the specimen, modern ultrasonography can reveal the vascular component in the mass before surgery and assist in diagnosis and surgical planning.[2]

Because the reproductive system is sensitive to radiation, computed tomography scans are seldom used for inspection of testicular pathological changes, while color Doppler ultrasound is a helpful diagnostic tool.[7] In the present case, the latter was used to identify the nature of the mass and to differentiate it from other testicular neoplasms. The report from the pathologist verified the mass in the right scrotum as benign testicular HLA and no testicular parenchyma was found, so we chose complete resection as the treatment method.

HLA is extremely rare in the scrotum.[6] Histological examination of the resected specimen showed both hemangiomatous and lymphangiomatous components. Necrosis and mitotic activity were absent. Although the etiology and pathogenesis of HLA have not been fully delineated, the coexistence of these two pathological entities may be related to abnormal development of the jugular lymphatic sacs in the embryo.[8]

In conclusion, our findings indicate that HLA is an extremely rare malformation of the lymphatic system and blood vessels in the newborn testis. This case report provides evidence that HLA may occur in the testis of neonatal patients.

Conflict of interest

The authors declare that there are no conflicts of interest.

Acknowledgments

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