Hemangiomas are benign vascular malformations of enlarged dysplastic vascular channels with abnormal growth of the endothelial cells. They are classified into capillary, cavernous, arteriovenous, venous and mixed subtypes. Cavernous and mixed are the most common types. Hemangioma is the most prevalent vascular tumor of childhood, affecting 1–3% of neonates and 10% of children by the age of 1 year [1]. Females are affected three to five times more frequently than males [2]. The most commonly involved sites are the musculoskeletal system, liver and spleen. About 2% of hemangiomas involve the genitalia and most of them are cutaneous lesions affecting females [3]. Deep hemangiomas of the scrotum are rare and important causes of scrotal mass in children. Hemangiomas are characterized by rapid growth, spontaneous regression and no recurrence [1–3]. The majority of hemangiomas present within the first two decades of life, and may enlarge during the growth of the child. Hemangiomas may be superficial (cutaneous), deep (subcutaneous), or may have both superficial and deep involvement [1]. History of a rapidly growing lesion in early childhood should arouse clinical suspicion. There may also be dilated veins and telangiectases [1]. Deep hemangioma may have a “bag of worms” feeling, reminiscent of a varicocele, but slightly firmer [1]. Ultrasonography is helpful but rarely definitive and can reveal these lesions [1]. Genital hemangioma involving the entire penis and scrotum is extremely rare. We have met several cases of scrotal hemangioma in our literature research. However, to our knowledge, this is the first scrotal hemangioma case in a patient older than 30 years of age. We present a large adult scrotal hemangioma and discuss the diagnostic and therapeutic problems.

CASE PRESENTATION

A 44-year-old man was admitted complaining of a scrotal mass involving the dorsal skin of the penis and
extending to the perineum. The mass gave the patient a dragging sensation and discomfort. On physical examination, we found a scrotal mass in the left side of the scrotum passing beyond the raphe in some areas, involving the dorsal skin of the penis and extending into the perineum. The patient reported that the scrotal mass had appeared in childhood with a very small size, and the lesions had enlarged slowly over time. The tumor presented as a soft, spongy, non-pulsatile, non-tender, irregularly lobulated mass measuring 18 × 8 cm, and was not attached to the testis (Figure 1A). Other clinical findings were normal. Laboratory data, including complete white blood cell counts, α-fetoprotein and human chorionic gonadotropin were normal. Bilateral testicular sonography was performed before surgical intervention. Subcutaneous tissue thickening and vein tortuosity were observed on scrotal color Doppler sonography exam, and the vascular tissue venous stream was determined. The patient underwent a direct scrotal approach. The mass was dissected and found to be a nodular, dark blue conglomerate of distended blood vessels measuring 16 × 4 × 4 cm, and was not attached to the testis or the spermatic cord, but was connected to the perineum by a pedicle containing a relatively large vein (Figure 1B). After dissection of the whole tumor, intact tissue of the scrotum, perineum and penis was restored (Figure 1C). Pathologic examination revealed a cavernous hemangioma (Figure 2). At the 16-month postoperative follow-up, the patient was healthy and free of complications and recurrence (Figure 1D).

**DISCUSSION**

Cavernous hemangioma, a subtype of hemangioma, may occur in mesenchymal tissue in any part of the body. Most hemangiomas are superficial lesions and have a predilection for the head and neck region. Cavernous hemangiomas are composed of large, dilated, blood-filled vessels lined by flattened endothelium. Several syndromes may be associated with cavernous hemangioma. Kasabach-Merritt syndrome, which includes thrombocytopenic purpura, is complicated with giant hemangioma [4]. A distinctive form
of a cavernous hemangioma of the skin, associated with similar gastrointestinal lesions, is blue rubber nevus syndrome [5]. Scrotal hemangiomas are rare entities. Proliferation goes on throughout the 1st year of life and is followed by slow involution. Thirty percent of hemangiomas resolve by 3 years, half by 5 years and 70–90% by 7 years [1]. The interesting point of our case is the age. Our patient was a 44-year-old male. As far as we know, this is the first case in which the patient is older than 30 years in the literature. One adult case was reported by Lent et al [6] in 1980. The case was a 30-year-old male with a large scrotal cavernous hemangioma while our case is a 44-year-old male with large scrotal hemangioma. Another case reported by Rastogi [7] was an 18-year-old man with hemangioma of the penis, scrotum, perineum and rectum. Nouira et al [8] reported an exceptional case of megapenis associated with corpus spongiosum agenesis and multiple hemangiomas of the scrotum, perineum and pelvis. Color Doppler sonography may demonstrate blood flow within these lesions but the absence of flow does not rule out the presence of these lesions. In our case, the vascular tissue venous stream was determined in color Doppler sonography. There was no testicular involvement in our patient and there was no erectile dysfunction or fertility problem. His sexual function was evaluated with the Florida Sexual History questionnaire. When the diagnosis is established, eradication of the lesion is recommended. Treatment options include a sclerosing agent (such as alcohol), surgery or laser therapy. Surgery and other treatment modalities are not always satisfactory, yield similar or less efficient results, and surgery has higher complication rates (e.g. scarring, deformity, erectile dysfunction, intraoperative bleeding or recurrence).

In conclusion, cavernous hemangioma involving the penis and scrotum is rare and may be associated with extension into the perineum, rectum and sigmoid colon, which may cause diagnostic and treatment difficulties. Imaging studies can determine the extent of the lesion, delineate its relationship with adjacent structures, and help plan therapy and follow-up.

REFERENCES


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