

Case Report

Bilateral Leiomyoma of Epididymides

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Leiomyoma is a relatively uncommon neoplasm of the epididymis derived from smooth muscle cells. A case of primary leiomyomas simultaneously occurring in both epididymides is reported in a 58-year-old man, clinically presenting painless intra-scrotal masses. The examination of the epididymis characterizes clinically by a firm solid, well-circumscribe mass, and histological spindle cells arranged in interlacing bundles with varying admixtures of fibrous and hyalinized connective tissue. Clinical, histopathologic and immunohistopathologic features, with briefly reviewed relevant literatures describing 7 cases of bilateral leiomyoma of epididymides are discussed. The patients with the mean age of 56.6 years (range 47-62 years) had epididymal mass measuring 5 to 75 millimeters (mean 21.3 millimeters). This is the first reported description of bilateral leiomyoma of epididymides in Thailand.

Keywords: Leiomyoma, Epididymis, Smooth muscle tumor, Bilateral

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Primary epididymal tumors are relatively uncommon^(1,2). Adenomatoid tumor is the most common diagnosed benign tumor of the epididymis, while leiomyoma is the second most common benign neoplasm of the epididymis⁽¹⁻⁴⁾. Characteristic clinical manifestation of epididymal leiomyoma is slow growing, painless intra-scrotal mass. It is occasionally mistaken clinically for malignancy. This report describes a patient with bilateral epididymal masses. Clinical investigations, histopathologic and immunohistopathologic features of both epididymides show characteristic findings of the leiomyoma.

Case Report

A married, 58-year-old Thai male patient living in Nontaburi, presented with bilateral painless firm scrotal masses of ten years' duration. The patient had no history of significant illness in the past. Palpation of the scrotum revealed a firm solid, well-demarcated nodule located at the head of each epididymis.

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The right and left nodules measured 2.5 and 1.5 cm in greatest dimension, respectively. No adhesion of the tumor to scrotal skin was noted. No lymphadenopathy was present. The patient underwent exploratory partial epididymectomy. Postoperatively, the patient recovered uneventfully.

The pathological examinations of the right and left epididymides showed well-defined, firm, tan-brown masses measuring 2.5 x 1.5 x 1.3 cm and 1.5 x 1 x 1 cm, respectively. The cut surfaces of both epididymides showed oval, rubbery, whorling, and white masses. The histopathologic feature revealed smooth muscle spindle cells arranged in interlacing bundles with varying admixtures of fibrous, and hyalinized connective tissue (Fig. 1, 2). No mitotic figure was detected in the spindle tumor cell. The adjacent epididymis was dilated. Immunohistopathologic investigations on the sections of both epididymal masses revealed positivity of smooth muscle antigen and vimentin in the spindle tumor cells. The cytokeratin (CK) epithelial membrane antigen (EMA), S100 and CD117 stains showed negative results in both epididymal tumors.

Discussion

Epididymal leiomyoma (EL) is a benign smooth

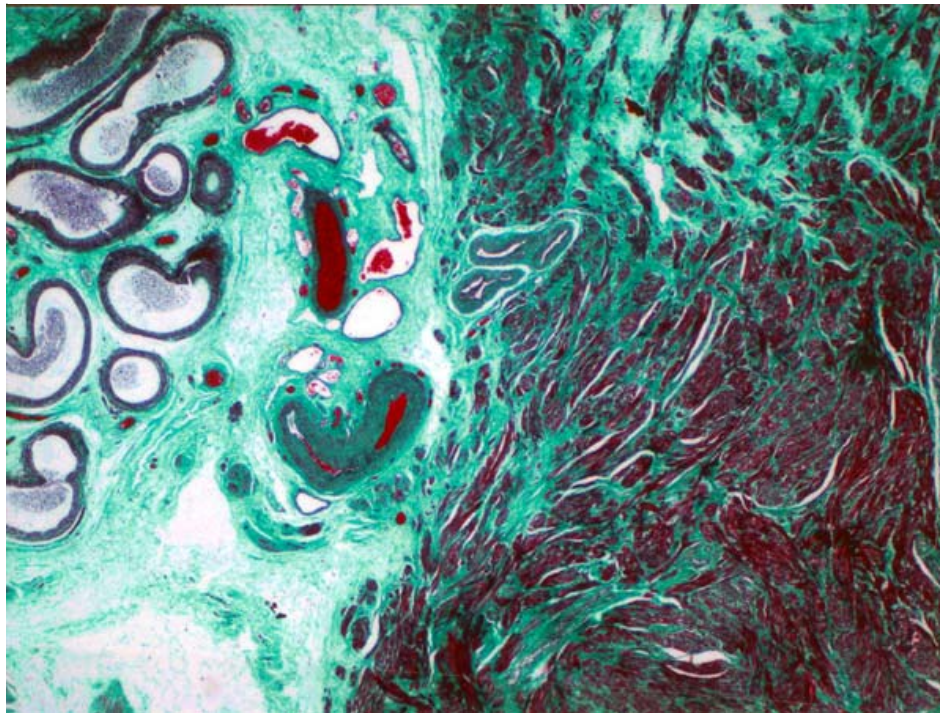


Fig. 1 The section of epididymis reveals smooth muscle spindle cells arranged in interlacing bundles with varying admixtures of fibrous, and hyalinized connective tissue. MASSONG trichrome, X20

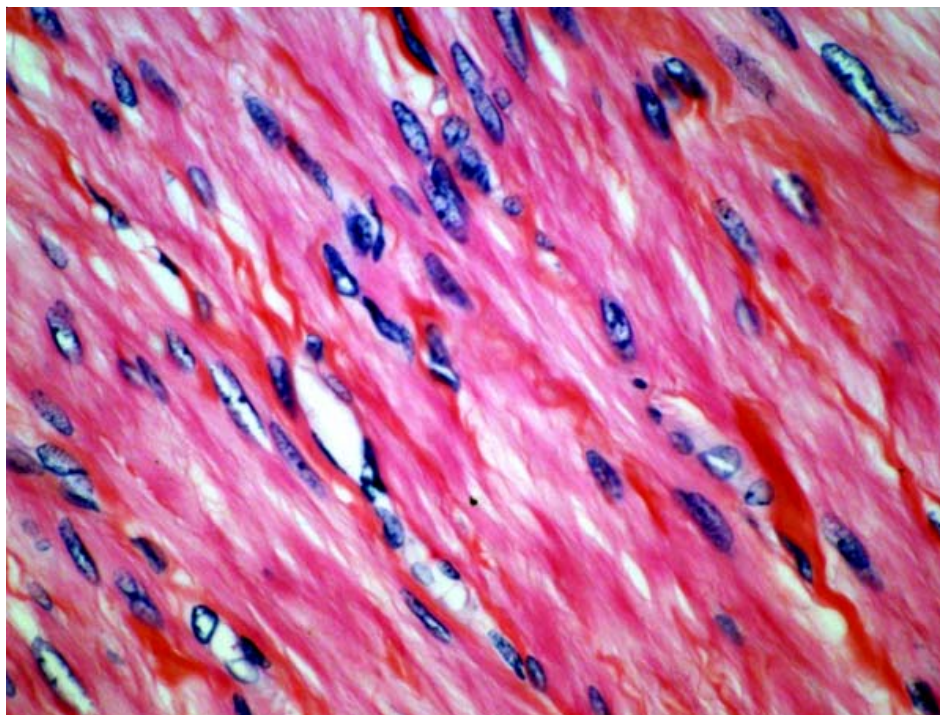


Fig. 2 The section of epididymis reveals spindle cells with cigar-shaped nuclei in leiomyoma. H&E, X400

muscle tumor, comprising 6% of epididymal tumors⁽⁴⁾. In Japanese literature, the incidence has been reported as high as 40% of the epididymal tumor⁽⁵⁾. It often cannot be distinguished from the other epididymal tumors including adenomatoid tumor, fibroma, lipoma, hemangioma, epididymal papillary cystadenoma, and malignant neoplasms, clinically⁽³⁾. EL is, however, usually unilateral, with one-fifth of the cases present with bilateral lesions⁽¹⁰⁾. The reported ages of patients with bilateral EL range from 47 to 62 years with a mean age of 56.6 years (Table 1). The published age at presentation of the unilateral EL occur during the second to ninth decade with a mean age of 51 years^(4,12). The mean ages of the onset of bilateral EL are slightly older than those with unilateral EL. Most patients presented with slow-growing, nontender epididymal mass⁽³⁾. Pain and discomfort are rare and are more often associated with malignant tumors⁽¹³⁾. EL typically produces a well-defined, firm mass surrounding with a gray-white fibrous capsule. The cut surface bulges and exhibits a whorled pattern. Tumor size ranges vary from 5 to 75 mm (mean 21.3 mm)⁽⁶⁻¹¹⁾. It frequently occurs in the globus minor or major and rarely in the corpus⁽⁴⁾. The histopathologic features show smooth muscle spindle cells arranged in interlacing bundles with varying admixtures of fibrous, often hyalinized connective tissue. Immunohistochemical studies of the spindle cell population in leiomyoma show the presence of smooth muscle antigen.

The clinical differential diagnoses of epididymal mass include tuberculous epididymitis, adenomatoid tumor, fibroma, lipoma, hemangioma, epididymal papillary cystadenoma, and leiomyosarcoma. Tuberculous epididymitis typically shows caseous granulomatous inflammation with positive organism by acid

fast staining method. Adenomatoid tumor is a benign mesothelial cell tumor characterized by numerous gland-like spaces, tubules or cords^(1,2). Lipoma is typically composed of mature adipocytes without atypia^(1,2). Hemangioma consists of dilated, thin-walled, mature vessels^(1,2). Epididymal papillary cystadenoma discloses tubules and cysts that often contain an eosinophilic colloid-like secretion with variable prominent papillae lined by cytologically benign columnar cells with clear, glycogen-rich cytoplasm project into the cysts^(1,2). Leiomyosarcoma characteristically show spindle cells forming compact fascicles with pleomorphic and hyperchromatic nuclei and multiple mitoses^(1,2,14). All histopathologic features of the clinical differential diagnosis of EL were not found in this case. Although, fibroma is typically composed of spindle, oval or round cells, it produces collagen that has similar histopathology to EL^(1,2). Positive results of immunohistochemical stains for smooth-muscle acting may be helpful in excluding fibroma. Finally, the definite diagnoses of bilateral epididymal tumors are leiomyoma.

Regarding the relatively uncommon site of a leiomyoma and clinical diagnosis is difficult in such a case. Therefore, the definite diagnosis of this condition should be confirmed by immunohistopathologic examination. Surgical excision is the most appropriate treatment. Resection of the tumor must be complete because some publishers suggest that leiomyosarcoma may arise from pre-existing leiomyoma^(14,15).

The present report describes the leiomyoma of bilateral epididymides in a 58 year-old man of a clinically, macroscopically, histopathologically and immunohistopathologically identical tumor. The authors believe that this is the first reported description in the literature of Thailand.

Table 1. Summary of 7 reported cases of bilateral leiomyoma of the epididymides

Authors & Year	Age (years)	Presented symptom	Tumor size (mm)		Location
			Right	Left	
Milner WA et al, 1939 ⁽⁶⁾	60	Painless masses	40	75	NA
Henderson ID et al, 1956 ⁽⁷⁾	47	Painless masses	15	6	Tail
Hata R et al, 1989 ⁽⁸⁾	50	Painless masses	5	10	Tail
Bruno S et al, 1993 ⁽⁹⁾	58	Hydrocele	15	25	Tail
Imazu T et al, 1994 ⁽¹⁰⁾	62	Painless masses	10	40	Tail
Kimura T et al, 1998 ⁽¹¹⁾	61	Painless masses	5	12	Tail
Larbcharoensub N et al	58	Painless masses	25	15	Head
Mean	56.6 yrs	-	21.3 (mm)		-

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รายงานผู้ป่วยที่มีเนื้องอกของกล้ามเนื้อเรียบบริเวณหลอดเก็บอสุจิทั้งสองข้าง

นพดล ลาภเจริญทรัพย์, พัฒนา ศรีมยุรา, วัชรินทร์ อริยประกาย

รายงานผู้ป่วยเนื้องอกปฐมภูมิของกล้ามเนื้อเรียบบริเวณหลอดเก็บอสุจิทั้งสองข้าง ในผู้ป่วยชายไทยอายุ 58 ปี คลำได้ก้อนที่บริเวณด้านในของถุงอัณฑะส่วนบน ตรวจทางกล้องจุลทรรศน์ และอิมมูโนฮิสโตเคมีพบเป็นเนื้องอกของกลุ่มเซลล์ของกล้ามเนื้อเรียบ ทางคณะผู้เรียบเรียงได้รายงานเรื่องเนื้องอกของกล้ามเนื้อเรียบบริเวณหลอดเก็บอสุจิทั้งสองข้างพบเป็นกรณีศึกษาแรกของประเทศไทย ร่วมกับบททบทวนจดหมายเหตุการแพทย์ บรรยายลักษณะเนื้องอกปฐมภูมิของกล้ามเนื้อเรียบบริเวณหลอดเก็บอสุจิทั้งสองข้างในผู้ป่วย 7 ราย ผู้ป่วยมีอายุเฉลี่ย 56.6 ปี (อายุระหว่าง 47 ถึง 62 ปี) โดยพบก้อนเนื้องอกขนาด 5 ถึง 75 มิลลิเมตร (ขนาดเฉลี่ย 21.3 มิลลิเมตร) โดยรวบรวมวิเคราะห์การแสดงออกทางคลินิกและพยาธิวิทยา