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Smooth Muscle Hyperplasia of the Epididymis
– Report of A Case and Review of the Literature –

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A 66-year-old man underwent surgery to remove an incidentally discovered non-tender intrascrotal mass. Ultrasonography revealed an irregular-margined, heterogeneous mass-like lesion in the epididymal tail. The mass was relatively well circumscribed but unencapsulated, irregular and firm; it consisted of expansile, increased smooth muscle fascicles originating from the epididymal muscular coat. Its cellular growth pattern lacked the cohesive, well-circumscribed proliferation pattern typical of a leiomyoma. A diagnosis of smooth muscle hyperplasia of the epididymis was made. Although ultrasonography is the imaging modality of choice for evaluating suspected intrascrotal masses, there are times when it cannot reliably identify the character of the masses and distinguish malignant from benign lesions. Ill-defined, solid extratesticular masses, that are ultrasonographically ambiguous, should be excised and confirmed histopathologically and smooth muscle hyperplasia of the epididymis should be included in the differential diagnosis of solid extratesticular masses.

Key Words : Epididymis; Smooth muscle; Hyperplasia

Intrascrotal masses can be divided into intratesticular and extratesticular lesions.[1] It is important to distinguish intratesticular from extratesticular masses, because most intratesticular masses are malignant, while extratesticular masses are usually benign.[1] In clinical practice, however, it may be difficult to make the determination preoperatively, so all intrascrotal masses should be appropriately evaluated for the possibility of a malignancy. Although ultrasonography is an accurate way to localize the lesions, and it is considered the imaging modality of choice for evaluating suspected intrascrotal masses, sometimes the ultrasonography cannot reliably identify the character of solid extratesticular masses.[1,2] When the origin or the character of the lesion is unclear after ultrasonography, the best course of action is surgical exploration followed by histopathological evaluation, as this is the only way to determine the specific diagnosis.[3]

Smooth muscle hyperplasia of the testicular adnexa is extremely rare. Because of the anatomical and histological complexity of the paratestis and scrotum, it is also difficult to identify the origin of smooth muscle proliferation. The epididymis, the abnormal overgrowth of smooth muscle is extremely rare, especially in the epididymis, and it has not been well studied.[4] Smooth muscle hyperplasia of the epididymis can mimic primary epididymal neoplasms such as leiomyoma or leiomyosarcoma or chronic epididymitis; it can also be overlooked during ultrasonography altogether. Here we report a rare case of smooth muscle hyperplasia originating from the epididymal muscular coat.

CASE REPORT

A 66-year-old man with a history of a low anterior resection for colon cancer 5 years earlier and of an inguinal lipoma 15 years before that presented with pain and a tingling sensation in the left inguinal region. Physical examination revealed a 3-cm soft, movable, nontender mass in the left inguinal region and an incidentally discovered ill-defined, nontender mass in the left scrotum. No adhesion of the mass to the scrotal wall was observed, and both testicles and the right epididymis appeared normal.

The serum level of tumor markers including CA 19-9 (15.26 U/mL) and carcinoembryonic antigen (1.9 ng/mL), were all within normal limits. Ultrasonography revealed a well-defined, homogenous soft tissue mass in the left inguinal region and an irregular-margined, heterogeneous mass-like lesion in the left epididymal tail (Fig. 1). The inguinal mass was consistent with a lipoma, but the intrascrotal mass was difficult to characterize. A small hydrocele was identified in both testicles, but the mass itself did not appear to involve the left testicle. A clinical diagnosis of recurrent inguinal lipoma and chronic epididymitis was made. An excision of the inguinal mass and an epididymectomy were performed. There was a relatively well-circumscribed, firm mass posterolateral to the epididymal tail, and it appeared to be easily dissected from the epididymis. The left testicle was unremarkable. Examination revealed that the inguinal mass was a 3 × 2.5 × 2-cm ovoid, yellow/brown, soft mass with a thin fibrous capsule and a homogenous cut surface; the epididymal mass was a 3.5 × 3 × 1.5-cm relatively well-circumscribed but unencapsulated, dark brown bit of firm muscular tissue. The proximal vas segment showed irregular thickening and tortuosity. Microscopic examination revealed that the inguinal mass was well-circumscribed and composed of mature adipocytes with fibrous septae. There was no cytological atypia, degeneration or necrosis. The mass was classified as a lipoma. The scanning view of the epididymal mass showed a solid, expansile lesion that continued into the epididymal muscular coat dissected from the parenchyma (Fig. 2). The lesion consisted of increased smooth muscle fascicles arranged in nodular patterns together with variable-sized vascular channels and entrapped fat (Fig. 3). The cellular growth pattern lacked the cohesive, well-circumscribed proliferation typical of a leiomy-

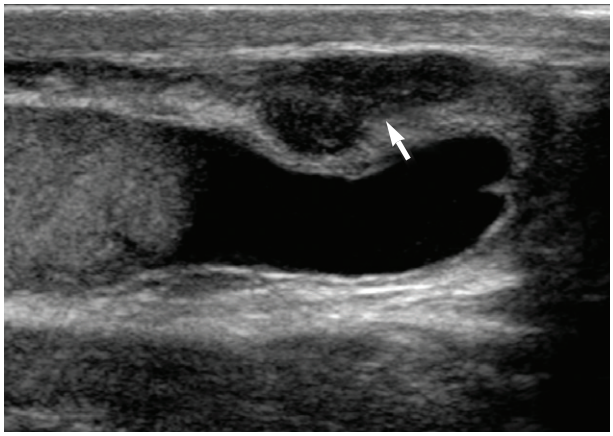

Fig. 1. Scrotal ultrasonography shows an irregular-margined, heterogeneous mass (arrow) in the left epididymal tail, measuring about 1.7 cm in diameter. A small hydrocele is also noted.

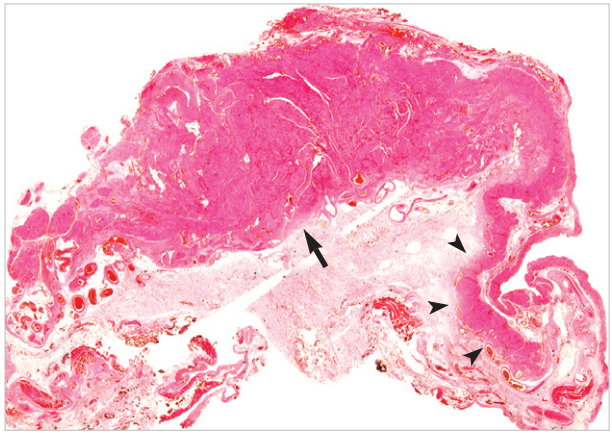

Fig. 2. Scanning view shows a relatively well circumscribed but unencapsulated, expansile mass (arrow) continued to the epididymal muscular coat (arrowheads) dissected from the parenchyma.

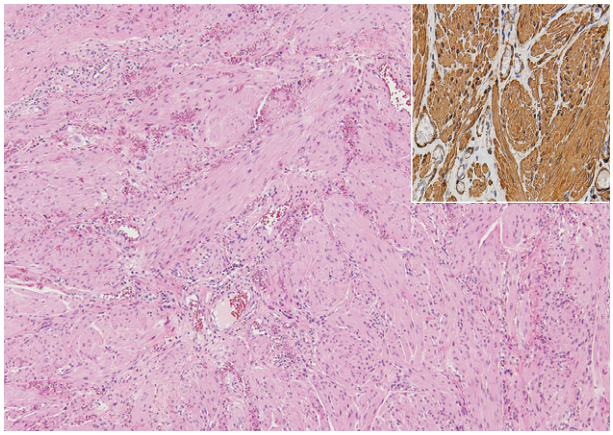

Fig. 3. There is an increase in smooth muscle fascicles arranged in a nodular pattern. Those fascicles are immunoreactive for smooth muscle actin (inset).

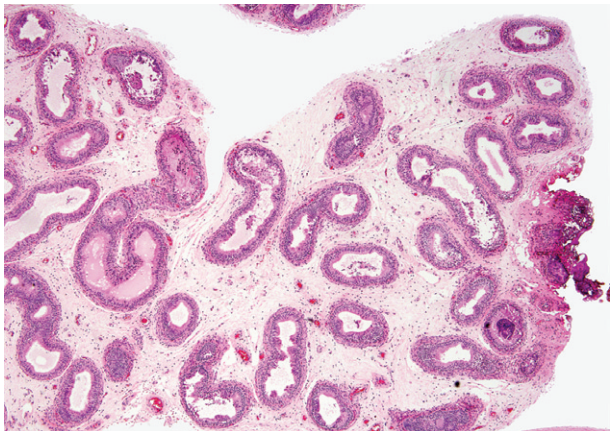

Fig. 4. Denuded epididymal parenchyma showed mild ductal atrophy without periductal or interstitial smooth muscle proliferation.

oma. There was occasional nuclear pleomorphism, but no mitotic figures or necrosis was identified. Immunohistochemical analysis revealed the smooth muscle phenotype of spindle cells, which displayed smooth muscle actin immunoreactivity (Fig. 3, inset). The remaining epididymal parenchyma had atrophic epididymal ducts (Fig. 4). However, there was no periductal or interstitial smooth muscle proliferation in the parenchyma. In addition, a tortuous vas segment and caudal epididymal ducts were surrounded by an orderly, concentric proliferation of smooth muscle. Thus a diagnosis of smooth muscle hyperplasia originating from the epididymal muscular coat was made. Postoperatively, the patient recovered well and, eight months later, showed no sign of recurrence.

DISCUSSION

When a scrotal mass is evaluated, the two most important questions are 1) whether the mass is intratesticular or extratesticular and 2) whether it is solid or cystic.[5] Solid intratesticular masses should be considered malignant, and cystic extratesticular masses are almost always benign. However, there is considerable overlap in the ultrasonographic appearance of solid extratesticular masses. Scrotal ultrasonography is generally considered to be accurate for localizing intrascrotal lesions, but its role in evaluating solid extratesticular masses is limited.

Non-neoplastic smooth muscle proliferation rarely occurs in the testicular adnexa.[6] This underrecognized lesion has been poorly described in the literature and is labeled with a variety of terms, including smooth muscle hyperplasia, smooth muscle hamartoma, leiomyoma and vascular leiomyoma.[6] Because of its rarity and the differences in the terms used to describe it, the clinicopathological features and pathogenesis of this smooth muscle proliferation have not yet been established. We found only 4 articles describing smooth muscle hyperplasia of the epididymis, including an original article and 3 case reports.[3,4,7,8] Barton *et al.* collected 16 cases of an analogous lesion in the testicular adnexa that clinically mimicked a neoplasm, and they reported that these cases had smooth muscle proliferation without the microscopic features of neoplasia.[4] Nine of the 16 cases were located in the epididymis, and the clinical diagnoses were epididymitis or an epididymal mass in 8 of the cases. In all cases microscopic examination showed

Table 1. The clincopathological features of five cases with smooth muscle hyperplasia that developed from the epididymis

	Payan *et al.*[8] (1967)	Payan *et al.*[8] (1967)	Sun *et al.*[7] (2003)	Kikugawa *et al.*[3] (2003)	This case (2008)
Clinical features					
Age (years)	55	70	55	76	66
Location	Both	Left	Right, tail	Left, head	Left, tail
Size (cm)	N/A	N/A	2.9×2.4	1.5	3.5×3
Presentation	Persistent scrotal pain	Persistent scrotal pain	Intrascrotal mass	Intrascrotal mass	Incidental
Physical exam	Scrotal enlargement	Cystic, tender mass	Paratesticular rubbery mass	Well-demarcated, non-tender mass	Ill-defined, non-tender mass
Medical history	Inguinal hernia Hydrocele	Hydrocele	Diabetes mellitus Hepatitis C	N/A	Inguinal lipoma Colon cancer
Ultrasonography	Within normal limits	Within normal limits	Paratesticular mass involving testis	N/A	Irregular-margined, heterogeneous lesion
Clinical diagnosis	Hydrocele and epididymo-orchitis	Chronic epididymo-orchitis	Paratesticular mass	Leiomyoma	Chronic epididymitis
Operation	Epididymo-orchiectomy	Epididymo-orchiectomy	Radical orchiectomy	Epididymectomy	Epididymectomy
Recurrence	N/A	No	N/A	No, during 22 months	No, during 8 months
Pathological features					
Gross finding	N/A	N/A	Vaguely-circumscribed, firm and tan/white	N/A	Relatively well-circumscribed, firm and dark brown
Growth pattern	Periductal	Periductal	Periductal Perivascular	Periducta Perivascularl	Nodular
Additional findings	Ductular dilatation and constriction	Ductular dilatation and constriction	Testicular involvement	Micro-tumors around main tumor	Muscular coat origin
Pathological diagnosis	DLP	DLP	SMH	SMH	SMH

N/A, not available; DLP, diffuse leiomyomatous proliferation; SMH, smooth muscle hyperplasia.

an increase in the smooth muscle fascicles arranged in a periductal, perivascular, or interstitial pattern and a lack of the cohesive, well-circumscribed proliferation of interlacing smooth muscle fascicles typical of a leiomyoma.[4] In 1967 Payan *et al.* reported two cases with diffuse smooth muscle proliferation in and around the epididymal ductules, which were diagnosed as "diffuse leiomyomatous proliferation of epididymis".[8] In those cases, the proliferation of smooth muscle bundles involving the epididymal parenchyma caused a constriction or dilatation of the ductules, and it included a hydrocele sac in one case. The authors suggested that, although multiple microscopic leiomyomata were present in the epididymal parenchyma, the etiology appeared to be smooth muscle proliferation rather than neoplasia since the pattern was diffuse and ill-defined.[8] In 2003 Sun *et al.*[7] reported a case of smooth muscle hyperplasia involving the parenchyma of the epididymal tail that focally extended into the inferior pole of the testis. Kikugawa *et al.*[3] also reported a case of smooth muscle hyperplasia of the epididymal head.

The clinicopathological features of 5 cases, including this case and 4 cases previously published in the English literature, are summarized in Table 1. The 16 cases reported by Barton *et al.* are not included because they did not report the origin of each case. The patients in 5 cases ranged in age from 55 to 76 years (mean 64.4 years) at the time of diagnosis. There was no predilection for the left or right side, and the the masses were found on both sides in 1 case. In the 3 cases that reported the size, the mass ranged from 1.5 to 3.5 cm (mean, 2.6 cm) in the greatest dimension. The clinical presentation included scrotal pain or discomfort (3/5) and palpation of intrascrotal masses (2/5). The clinical diagnoses included chronic epididymo-orchitis (2/5), chronic epididymitis (1/5), a paratesticular mass (1/5), leiomyoma (1/5), and hydrocele (1/5). All five patients underwent surgery including an epididymectomy (2/5), epididymo-orchiectomy (2/5) and radical orchiectomy (1/5). In the 3 cases that reported the location, the masses were located in the tail (2/5) and the head (1/5) of the epididymis. In the 4 cases previously reported, the histological examination revealed increased smooth muscle fascicles arranged in mixed perivascular, interstitial or periductal patterns without cellular cohesiveness; the main pattern was periductal. Dilatation and constriction of the epididymal ductules was noted in three cases. In the case we are reporting, smooth muscle fascicles were arranged in nodular patterns, and periductal smooth muscle proliferation was absent in the epididymal parenchyma. Focal involvement of the testis was present in 1 case.[7] Follow up was documented in two patients. These two patients were free from recurrence for the duration of their follow-up periods, which lasted an average of 15 months.

The English literature contains reports of non-neoplastic or neoplastic smooth muscle proliferations in a number of different structures of the testicular adnexa and scrotum, including the tunica dartos, tunica albuginea, tunica vaginalis testis, and epididymis.[3,9-12] However, no report befor this one had determined the anatomical origin of the smooth muscle proliferation. A tunica dartos origin of the smooth muscle hyperplasia could be excluded in this case because the mass in the epididymal tail was located apart from the scrotal wall. The tunica albuginea and tunica vaginalis testis could also be excluded because the mass had no contact with the testis. The tunica albuginea contains some smooth muscle cells, concentrated mostly on the posterior aspect of the testis, and leiomyomas are thought to involve smooth muscle in the vascular tree of the tunica albuginea.[11,13] In this case, however, the mass was located posterolateral to the epididymal tail and had no angioleiomyomatous features. The histological differentiation between the epididymis and other paratesticular adnexa has not reported previously. The tunica vaginalis is composed of serous membrane and is covered with a single layer of simple cuboidal endothelial cells. We found no histological evidence of smooth muscle proliferation from the serous epithelium. We diagnosed the patient with smooth muscle hyperplasia according to following histopathological findings: 1) continuity of the lesion to the dissected muscular coat noted during the epididymectomy; 2) absence of contact with the testis and scrotum and no anatomical relation with the previous operation for inguinal lipoma or the low anterior resection for colon cancer; 3) noncohesive mass-forming appearance; and 4) variation in the thickness of the mass.

We have presented a 66-year-old man with smooth muscle hyperplasia that developed from epididymal muscular coat. We suggest that when ultrasonography reveals ill-defined, heterogeneous, solid mass-like lesions in the paratesticular region, smooth muscle hyperplasia should be included in the differential diagnosis.

REFERENCES

1. Frates MC, Benson CB, DiSalvo DN, Brown DL, Laing FC, Doubilet PM. Solid extratesticular masses evaluated with sonography: pathologic correlation. Radiology 1997; 204: 43-6.
2. Sung T, Riedlinger WF, Diamond DA, Chow JS. Solid extratesticular masses in children: radiographic and pathologic correlation. AJR Am J Roentgenol 2006; 186: 483-90.

3. Kikugawa T, Tanji N, Kurihara K, Shishido S, Mannami M. Smooth muscle hyperplasia of the epididymis: a case report. Pathology 2003; 35: 454-5.
4. Barton JH, Davis CJ Jr, Sesterhenn IA, Mostofi FK. Smooth muscle hyperplasia of the testicular adnexa clinically mimicking neoplasia: clinicopathologic study of sixteen cases. Am J Surg Pathol 1999; 23: 903-9.
5. Woodward PJ, Schwab CM, Sesterhenn IA. From the archives of the AFIP: extratesticular scrotal masses: radiologic-pathologic correlation. Radiographics 2003; 23: 215-40.
6. Nuciforo PG, Roncalli M. Pathologic quiz case: a scrotal sac mass incidentally discovered during autopsy. Arch Pathol Lab Med 2003; 127: 239-40.
7. Sun X, Gong Y, Pins MR. Pathologic quiz case: a paratesticular mass in a 55-year-old man. Arch Pathol Lab Med 2003; 127: E111-2.
8. Payan HM, Mendoza C. Diffuse leiomyomatous proliferation in the epididymis. Arch Surg 1967; 94: 427-9.
9. Forte F, Scarfini M, Spera E, *et al*. Leiomyoma of the epididymis and testicular adnexa: apropos of 3 cases. Arch Ital Urol Androl 2004; 76: 119-20.
10. Satyanarayana S, Jawed KZ, Sikri V, Kaur B, Singh P. Myxoid leiomyoma of tunica vaginalis testis. Indian J Pathol Microbiol 2001; 44: 373-4.
11. Mak CW, Tzeng WS, Chou CK, Chen CY, Chang JM, Tzeng CC. Leiomyoma arising from the tunica albuginea of the testis: sonographic findings. J Clin Ultrasound 2004; 32: 309-11.
12. Quinn TR, Young RH. Smooth-muscle hamartoma of the tunica dartos of the scrotum: report of a case. J Cutan Pathol 1997; 24: 322-6.
13. Dogra VS, Gottlieb RH, Oka M, Rubens DJ. Sonography of the scrotum. Radiology 2003; 227: 18-36.