A Case of a Cavernous Type of Angioleiomyoma Occurring on the Buttock

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Angioleiomyoma(ALM) of the cavernous type is a rare subtype of leiomyomas arising from the smooth muscle of veins. ALM of this type invariably shows clinically and histopathologically distinctive features, compared with the classical solid or venous type. However, no case of ALM of this type has been yet reported in Korea although there have been several reported cases of other types. We herein present the case of a 39-year-old man with a 7-year history of a painless ALM of the cavernous type on his buttock with the histological features of markedly ectatic vascular channels and thin intervascular bundles of smooth muscles. (Ann Dermatol 11(1) 33~36, 1999).

Key Words : Angioleiomyoma, Cavernous type.

Angioleiomyoma(ALM), a rare benign neoplasm showing differentiation towards the smooth muscle walls of veins, represents a painful solitary nodule and is most frequently reported on the lower extremities of middle-aged women[1-2]. However, this typical characterization is actually limited to tumors of the solid type according to Morimoto's classifying mode of ALMs[3]. Although a cavernous type is rarest among three histological subtypes of ALMs, it invariably shows clinically and histopathologically different features from other subtypes[3-6].

We report a case of the cavernous type of ALM occurring on the buttock of a 39-year-old man. Such a case has not been reported in the Korean literature.

CASE REPORT

A 39-year-old male had had a nonpainful, solitary skin lesion of 7 years' duration. An examination demonstrated a well-defined, dome-shaped, erythematous, soft nodule on his buttock. It was 3 cm in size at its greatest dimension(Fig. 1). An initial diagnosis of an epidermal cyst or dermatofibroma was made and his nodular lesion was surgically excised. A skin specimen taken from his skin lesion was processed for hematoxylin-eosin-stained sections. A skin biopsy specimen showed a well-circumscribed intradermal tumor which consisted of remarkably dilated, numerous vascular channels with little muscular thickening of the walls(Fig. 2, Fig. 3). There were smaller amounts of intervascular smooth muscle fibers in the tumor. Therefore, the muscular walls of these vessels were difficult to distinguish from intervascular smooth muscle bundles. The muscle fibers encircling the ectatic blood vessels contained blunt-tipped, straight and longer nuclei, and abundant eosinophilic cytoplasms(Fig. 4). There were no myxoid or hyaline changes. We could not find groups of mature fat cells in the tumor. The overlying epidermis was relatively intact. Masson's trichrome staining could separate smooth muscle bundles from the sparce strands of collagen fibers(not shown here). The results of immunohistochemical staining showed positivity to desmin, α -smooth muscle actin and vimentin but not to neuron specific

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Fig. 1. A well-defined, dome-shaped, erythematous, soft nodule on his left buttock region.

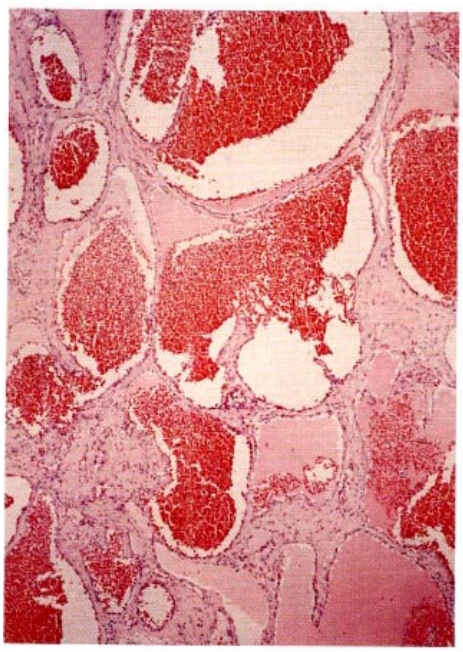


Fig. 3. Markedly dilated, numerous vascular channels with little muscular thickening of the walls(H & E, × 200).

enolase and S-100 antigen(Dako, Copenhagen, Denmark)(Fig. 5). The endothelia of numerous blood vessels had positivity to factor VIII-related antigens(Dako)(Fig. 5). At the time of writing, his

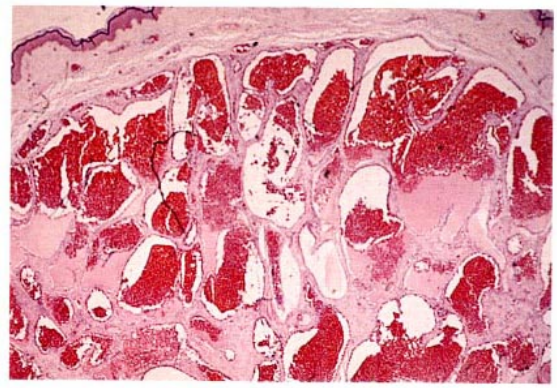


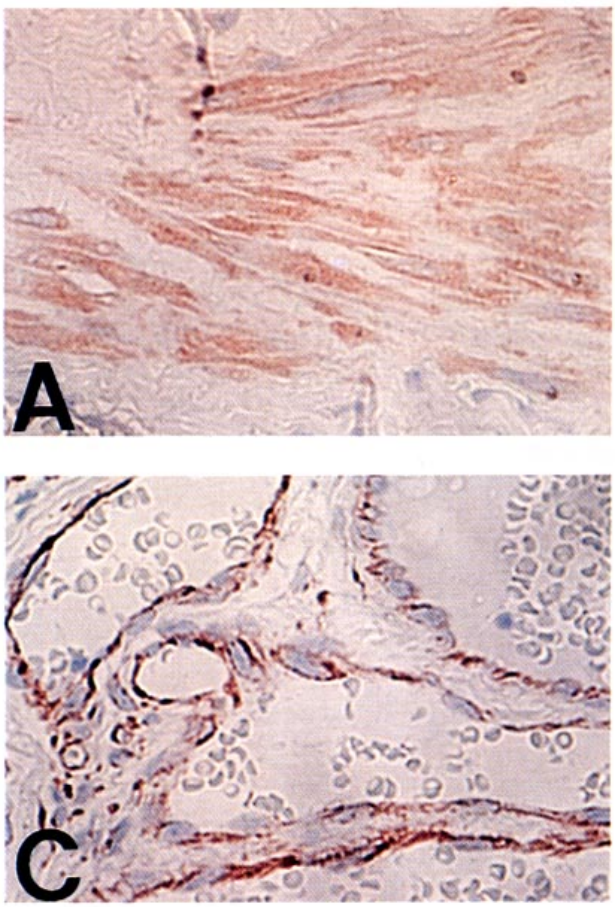
Fig. 2. A well-circumscribed intradermal tumor composed of numerous blood vessels(H & E, ×100).

Fig. 4. The muscle fibers encircling ectatic blood vessels contained blunt-tipped, straight and longer nuclei and abundant eosinophilic cytoplasms(H & E, ×400).

skin lesion had not recurred 8 months after surgical excision.

DISCUSSION

Five types of cutaneous leiomyomas have been defined as follows; 1) solitary or 2) multiple pilo-leiomyomas, arising from arrector pili muscles; 3) solitary genital leiomyomas, arising from the dartoic, vulvar, or mammillary muscles; 4) solitary



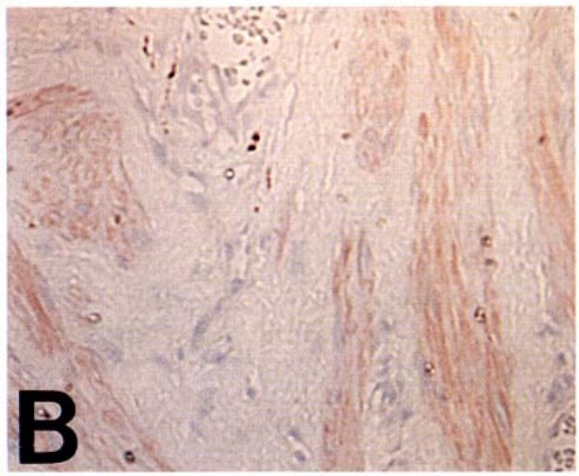


Fig. 5. Immunohistochemical findings of muscle fibers reacting with desmin(A, ×200) and α-smooth muscle actin(B, ×200). The endothelia of numerous blood vessels demonstrating the positivity to factor VIII-related antigens(C, ×200).

ALMs, arising from the muscles of veins; and 5) leiomyomas with additional mesenchymal elements. Additionally, there are three subtypes in solitary ALMs; a solid or capillary type, a venous type, and a cavernous type[1-6]. Although there were several reports of ALMs of the solid type, tumors of the cavernous type as in this case had not been yet reported in Korea untill now[7-11].

Clinically, a typical solid type, the most common(60-70%) subtype of ALMs, represents a painful, tender solitary nodule occurring upon the lower extremities of middle-aged women[3-5]. These tumors are generally quite smaller, most being less than 1 cm in their greatest dimension[4]. In the solid type, contraction of compact vascular muscles, distortion of the myelin sheath, and subsequent local ischemia probably give rise to unique lesional pain[1]. The consistency of these tumors is usually firm or hard[3]. However, a cavernous type which is the rarest(10-12%) subtype of ALMs, usually shows a 3-5 cm sized, painless nodule in the upper extremities or trunk of men[1,3]. Painfulness rarely occurs in cavernous type, because there is no local ischemia by muscular contraction or damage to nerve fibers in the tumor[3,5]. The consistency of tumors of cavernous type is relatively soft due to a large number of dilated vascular channels[3]. Our case of the cavernous type showed a 3 cm sized, painless, non-tender, soft nodule on the buttock of a 39-year-old man.

Histopathologically, all ALMs have common findings in that they are well-circumscribed and contain numerous vessels surrounded by interlacing bundles of smooth muscles that fuse with the vessel walls[4]. Regardless of the histological subtypes, the smooth muscle fascicle cells encircling blood vessels, contained centrally located, blunt-tipped, "eel-like", straight and longer nuclei, and abundantly eosinophilic and vacuolated cytoplasms[1,4]. However, according to histological subtypes, the numerous blood vessels that are present vary in size and have muscular walls of varying thickness. On this basis, three subtypes can be differentiated respectively. In the solid type, the vascular channels are numerous but small[5]. This type usually shows the vessels with a rounded or slit-like or stellate lumen because of contraction of their muscular tissues. It often reveals evidence of luminal occlusion or organization of thrombi[6]. The vascular spaces are typically surrounded by a closely compacted muscular coat arranged in orderly concentric layers

that blend into areas with proliferation of muscle fascicles[7-9]. In the venous type, the veins exhibit thick but not so compact muscular walls. Encircling smooth muscle cells extend tangentially from the peripheries of the veins and merge with the intervascular tumor substance[5]. Focal calcification, hyaline changes, or mucinous alterations are often present in venous type[3]. Finally, tumors of the cavernous type are composed of prominently dilated vascular channels with small amounts of smooth muscle[1]. This type invariably shows little muscular thickening of the walls and not so compact intervascular smooth muscles fibers as in the solid type[3,5,6]. Our case demonstrated the compatible findings with ALM in that the intradermal tumor was well-circumscribed and had numerous dilated vessels, interlacing bundles of intervascular smooth muscle, the cells of smooth muscle fascicles with centrally located, blunt-tipped, straight and longer non-wavy nuclei and little muscular thickening of the vessel walls.

Histopathological differentiation from several diseases should be considered; angioleiomyolipoma, arteriovenous(A-V) hemangioma, subcutaneous cavernous hemangioma, leiomyosarcoma, glomangioma, and glomangiomyoma. Recently, angioleiomyolipoma(or angiolipomyoma, angiolipoleiomyoma) can arguably be considered an ALM with fat cell modulation[1,12-14]. However, it has elastic lamina completely rimming the vascular channels and represents an asymptomatic subcutaneous tumor which is composed of smooth muscle, vascular spaces, connective tissue and mature fat. Because we could not find groups of mature fat cells in the tumor, the differential diagnosis from angioleiomyolipoma could be made. A-V hemangiomas consist of multiple dilated blood vessels in the dermis as in ALM. However, they invariably show thick-walled muscle-containing blood vessels, lined by a single layer of endothelial cells[1,15]. In subcutaneous cavernous hemangiomas, the vascular spaces are larger, the vessel walls are thinner, the tissues surrounding the vessels lack fascicles of smooth muscles, and the tumors lack a fibrous pseudocapsule[1].

Surgical excision is the mainstay of treatment for ALM[13]. In our case, there was no evidence of recurrence 8 months after surgical excision.

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