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BRIEF COMMUNICATION

Intranodal Palisaded Myofibroblastoma; a Case Report and Review of the Literature

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Abstract Intranodal palisaded myofibroblastoma (IPM) also called as intranodal hemorrhagic spindle cell tumor with amianthoid fibers is a distinctive and rare mesenchymal neoplasm of lymph nodes. This entity generally misdiagnosed as intranodal Kaposi's sarcoma or schwannoma in past. In contrast to Kaposi's sarcoma, it behaves in a benign fashion and does not need any further therapy except total surgical resection of the mass. This neoplasm has a great predilection for the inguinal region. The lesion presents typically as a unilateral, painless, solitary mass. To our knowledge, approximately 53 cases of IPM have been reported in the English-language literature. We present a 43-year-old-male patient with IPM and discuss histological, immunohistochemical features and pathogenesis of this rare benign neoplasm.

Keywords Myofibroblastoma · Intranodal palisaded myofibroblastoma · Amianthoid fiber

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Introduction

Intranodal palisaded myofibroblastoma (IPM) also called as intranodal hemorrhagic spindle cell tumor with amianthoid fiber is a rare benign mesenchymal neoplasm of lymph nodes. IPM was first described by different authors in 1989 (1–3). Inguinal lymph nodes are a primary originating site for IPM. Submandibular and cervical lymph nodes were also reported as rare originating sites (4–6). To our knowledge, approximately 53 cases of IPM have been reported in the English-language literature (1–20). The ages of patients ranged from 19 years to 71 years. The slight male predominance (M/F: 1.4) was present (Table 1).

Case History

A 43-year-old man who has been following up with the diagnosis of low-grade glial tumor presented with a swelling in his left groin. In physical examination, a solitary, painless, firm, mobile mass was determined in the left inguinal region of the patient. The mass was totally excised. Macroscopically, the specimen consisted of a firm and encapsulated mass, measuring $4.5 \times 3.5 \times 2.5$ cm. On cut sections, the lesion had a nodular, white-grey and brownish appearance with focal haemorrhage areas. The lesion was an appearance of a conglomerated mass of lymph nodes (Fig. 1).

Histopathological Findings

In the histological slides, a narrow rim of lymphoid tissue with marginal sinuses was observed. This finding pointed out that the lesion originated from a lymph node. Almost whole lymph node was involved by the lesion and a pseudo capsule

Table 1 The cases of intranodal palisaded myofibroblastoma in the English-language literature

Source	No. of cases	Age (year)	Sex (no)	Location
Weiss et al. [1]	22	26–67	M (15), F(7)	Inguinal
Suster and Rosai [2]	6	19–61	M (4), F(2)	Inguinal
Lee et al. [3]	1	48	F	Inguinal
Alguacil-Garcia [4]	1	61	M	Submandibular
Fletcher and Stirling [5]	2	35, 40	F (2)	Submandibular
Michal et al. [6]	6	47–63	M (2), F(4)	Inguinal(5), cervical(1)
Creager et al. [7]	1	49	F	Inguinal
Bigotti et al. [8]	1	51	M	Inguinal
Kleist et al. [10]	1	71	M	Inguinal
Lioe et al. [11]	1	51	F	Inguinal
Barbareschi et al. [12]	1	53	F	Inguinal
Tanda et al. [13]	1	70	F	Inguinal
Eyden et al. [14]	1	48	M	Inguinal
Rossi et al. [15]	3	42–67	M (2), F(1)	Inguinal
Burns et al. [16]	1	45	M	Inguinal
Onsurbe et al. [17]	1	56	M	Inguinal
Ciralik et al. [18]	1	48	F	Inguinal
Mies et al. [19]	1	52	M	Inguinal
Cobanoglu et al. [20]	1	60	M	Inguinal

separated the lesion from residual lymphoid tissue. The lesion was composed of bland looking spindle cells with areas of nuclear palisading in short intersecting fascicles similar to that of the Antoni-A pattern seen in schwannomas (Fig. 2). Nuclear palisading was focal and vague in the central of lesion (Fig. 3). Spindle cells characterized by scant cytoplasm with indistinct borders and elongated nuclei with coarse chromatin. Perinuclear vacuolization and mild nuclear pleomorphism were noticed in some spindle cells. There were no mitotic activity and necrosis. In many areas, especially beneath

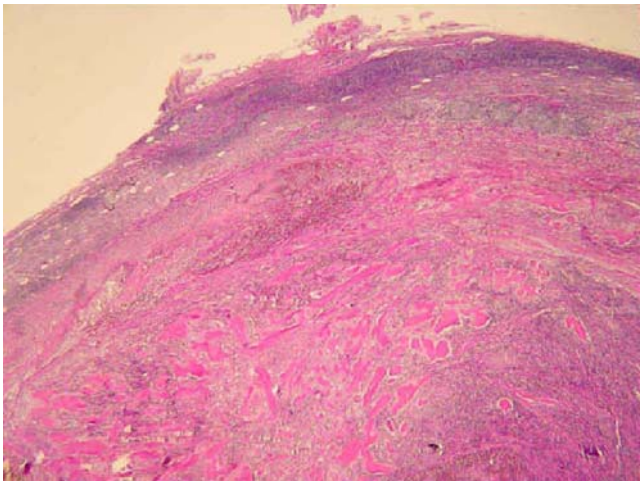

Fig. 2 Low magnification shows the rim of compressed lymph node in the periphery of the tumoral lesion (X10, H&E)

pseudo capsule, clefts forming by spindle cells filled by red blood cells were present. Fresh and old foci of hemorrhage were observed beneath the pseudo capsule. Pseudo capsule contained vessels with thickened wall. Extensive collagenization and stellate shaped collagenous bundles so called "amianthoid fiber" were a characteristic finding of the lesion (Fig. 4). Gomori-trichrome stain displayed the collagenous nature of these structures. Some collagenous bundles contained small capillary vessels (Fig. 5). The complete and incomplete forms of collageneous bundles were also present.

Immunohistochemical Findings

Spindle cells were positive for smooth muscle actin (SMA), muscle specific actin (MSA), vimentin, while negative for desmin, S-100, CD117 (c-kit), CEA, keratins, CD34, calpo-

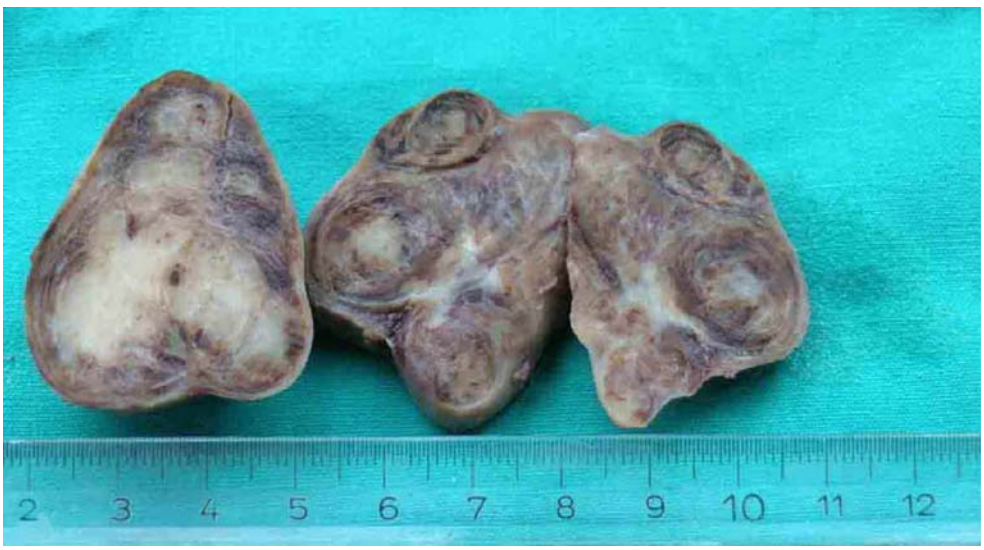

Fig. 1 The cut surface of well circumscribed tumoral lesion including hemorrhagic foci

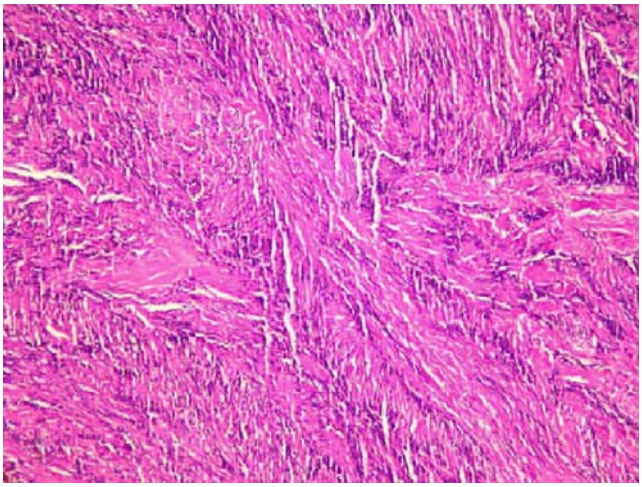

Fig. 3 Interlacing spindled cells showing nuclear palisading in the tumor (X50, H&E)

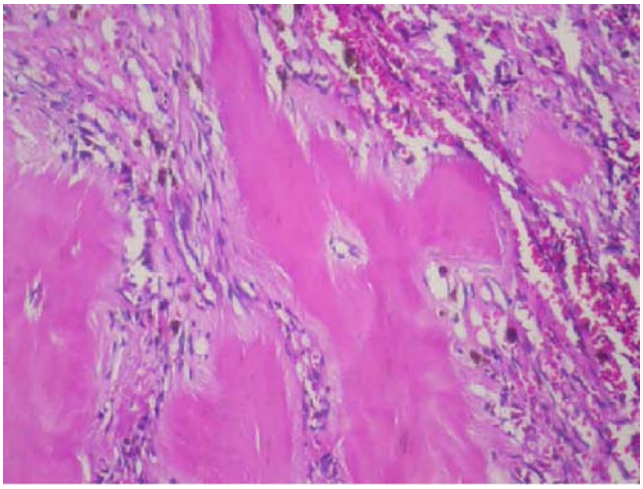

Fig. 5 A capillary vessel in the central portion of a collagenous bundle (X200, H&E)

nin, HMB-45, Epstein Barr virus latent mebran protein 1 (EBV LMP1), Herpes simplex virus type II (HSV-II) and Human papilloma virus (HPV). Positive staining for SMA and vimentin were determined especially around the amianthoid fibers. CyclinD1 expression was observed in the nuclei of approximately 10% of spindle cells.

On the basis of these histopathological and immunohistochemical findings, IPM originating from inguinal lymph nodes was diagnosed.

Discussion

Primary non-lymphoid neoplasms of lymph nodes are extremely uncommon. IPM showed some different morphological features than those of Kaposi's sarcoma and schwannoma. Bland nuclear atypia, large collagenous bundles, the absence of intra/extra cellular hyaline globules and negative immunostaining for CD34 were the distinguishing features from Kaposi's sarcoma, while negative staining for S-100 was a distinguishing feature from schwannoma [2, 4, 6]. Sophisticated studies pointed out to a myofibroblastic cell origin of IPM. Spindle cells of IPM were positive for SMA and vimentin but negative for S-100, Factor-VIII-related antigen, desmin, glial fibrillary acidic protein, epithelial membrane antigen and keratin antibodies [2, 7]. Spindle cells were characterized by a continuous expression of actin and vimentin and lack of desmin in all reports in the literature [8]. Our case had also a

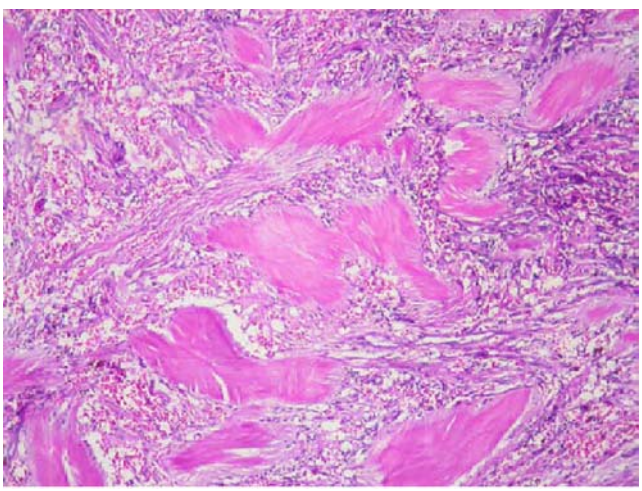

Fig. 4 Stellate shaped collagenous bundles (X50, H&E)

Table 2 The differential diagnosis of IPM

Differential diagnoses	Discriminative features from IPM
Kaposi sarcoma	High mitotic activity, nuclear pleomorphisim, CD34 positivity
Schwannoma	Coexistence of Antoni A and B, S-100 positivity
Leiomyosarcoma	High mitotic activity, nuclear pleomorphisim, necrosis, desmin, caldesmon, EMA and low molecular weight keratin positivity
Leiomyoma	Extensive whorls of smooth muscle cells, desmin, caldesmon, EMA and low molecular weight keratin positivity
Dentritic reticulum cell tumor (sarcoma)	Whorls of plump cells, the presence of small lymphocytes throughout the tumor cells (thymoma like appearance), CD21 positivity
Spindle cell melanoma	Nuclear pleomorphism, melanin pigment accumulation, HMB-45 positivity
Spindle cell carcinoma	Keratin positivity, foci or a small focus of squamous differentiation
Inflammatory myofibroblastic tumor	Inflammatory cell component, lower cellularity

similar immune profile. This immune profile was similar to that of certain normal vascular structures. A similar profile was also noticed in gastrointestinal stromal tumors of presumed smooth-muscle nature. C-kit immunostaining in IPM has not been reported until now. We have not also detected an immunostaining for c-kit. These distinctive tumors of lymph nodes might arise from the proliferation of desmin negative vascular smooth-muscle cells. The cells showing similar immunophenotype were determined in the wall of vessels locating in hilum and capsule of lymph nodes [2]. IPM arises from selectively inguinal lymph nodes. Bigotti et al. reported that actin positive/desmin negative myofibroblasts were diffusely prominent in inguinal lymph nodes compared with other lymph node regions. In addition, they suggested that the proliferation of myofibroblasts with this immunophenotype could be secondary to the increased drainage function in inguinal lymph nodes [8].

An overexpression for cyclinD1 (in approximately 50% of spindle cells) in IPM was reported in the literature [10, 19]. Kleist also performed a molecular analysis for cyclinD1 and found no any abnormal result. Kleist suggested that cyclinD1 played a role in promoting the growth of IPM due to its high expression, in the absence of genetic disturbances [10]. We did not find an overexpression for cyclinD1 in our case. The presence of EBV and human herpes virus-8 (HHV-8) genome in inflammatory myofibroblastic tumor was previously reported. Kleist et al. [10] and Creager et al. [7] also researched genomes of EBV and HHV-8 in IPM. But, they did not find any evidence for EBV and HHV-8 genomes. In our also case, immunohistochemical analysis performed for EBV LMP1, HSV-II and HPV failed to detect HSV, HPV and a latent EBV infection in IPM.

Collagenous bundles resembling amianthoid fibers were also described in some mesenchymal tumors [2, 6, 7]. Amianthoid fibers consist of abnormally thick collagen fibers. However, collagenous bundles of IPM were composed of morphologically normal collagen fibrils [9]. The collagenous nature of these structures in our case was determined by Gomori-trichrome stain. Immunohistochemically, type I and type III collagen were expressed throughout whole and in the peripheral areas of the structure, respectively [9, 10].

The differential diagnosis of IPM includes Kaposi's sarcoma, schwannoma, spindle cell carcinoma, spindle cell melanoma, leiomyoma, leiomyosarcoma, dendritic reticulum cell tumor and inflammatory myofibroblastic tumor (Table 2) [2, 6, 10].

IPM behaves in a benign fashion. In the literature, there were only two cases with local recurrence [7, 11]. Metastases have not been reported until today. The simple surgical resection is curative. Our patient is alive without evidence of disease, 3 years post-operatively.

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