CASE REPORT

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Multiple Spindle Cell Lipomas and Spindle Cell Lipoma as A Component of Multiple Lipomas: A Case Series of Seven Patients

Multiple İğsi Hücreli Lipomlar ve Multipl Lipomların Komponenti Olarak İğsi Hücreli Lipom: Yedi Olguluk Seri

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ABSTRACT

Spindle cell lipoma (SCL) is a benign lipomatous tumor consisting of mature adipose and spindle cells with a thick collagen component. Although SCLs are characteristically seen as solitary tumors in elderly male patients on the posterior neck, shoulder and back, they rarely can be presented as multiple tumors or synchronously with multiple ordinary lipomas in the same patient. It has been claimed that the natural progression of the disease might be sequential, and the later lesions were more cellular. We present 7 patients diagnosed as either multiple spindle cell lipomatosis or SCL as a component of multiple lipomas. In 4 tumors of two patients, we observed multifocal spindle cell proliferation areas within myxoid stroma containing ropy collagen bundles. The finding of SCL component as multiple small foci in otherwise ordinary lipoma-like mature adipocytic cells may support the claim that within a period, ordinary lipomas may be converted into SCLs.

Keywords: Spindle cell lipoma, lipoma, CD34

ÖZ

İğsi hücreli lipom (SCL) kalın kollagenöz komponent içeren matür adipöz ve iğsi hücrelerden oluşan benign lipomatöz bir tümördür. SCL'ler karakteristik olarak orta yaşlı erkek hastaların ense, omuz ve sırtında soliter olarak görülüyor olsa da nadiren multipl tümörler olarak ya da aynı hastada multipl olağan lipomlarla aynı anda bulunabilirler. Hastalığın doğal gidişinin sıralı olabileceği ve daha sonra gelişen lezyonların daha hücresel olabileceği iddia edilmiştir. Bu raporda multipl SCL veya multipl lipomların komponenti olarak SCL tanısı almış 7 hasta sunuldu. Hastalardan ikisinde toplam 4 lezyonda miksoid stroma içinde kalın, halatsı kollajen içeren multifokal iğsi hücre proliferasyonu gözlemlendi. İki hastada olağan lipom benzeri matür adipositik hücreler arasında multipl küçük SCL odaklarının varlığı bize göre belli bir zaman dilimi içinde olağan lipomların SCL'ye dönüşebileceği iddiasını destekliyor olabilir.

Anahtar Kelimeler: İğsi hücreli lipom, lipom, CD34

Introduction

Spindle cell lipoma (SCL) is a benign lipomatous tumor consisting of mature adipose and spindle cells with a thick collagen component, first described by Enzinger and Harvey (1) in 1975. Diagnosis can be difficult because of heterogeneous histopathological features (2,3,4). Although SCL is characteristically seen on the posterior neck, shoulder, and back as a solitary, well-circumscribed lesion in elderly men, multiple synchronous tumors can rarely be seen in a patient, and some of these are reported to be familial (5). SCL can rarely occur synchronously with multiple ordinary lipomas in the same patient (6,7,8). In this report, we presented seven patients with either multiple SCLs or SCL as a component of multiple lipomas.

Case Report

All patients were male. Clinicopathological features are summarized in Table 1.

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Table 1. Clinicopathological features

P	A	S	SCL (N)	L	Max diameter	Additional findings
1	47	M	5	Trunk	2.5 cm, 2.5 cm, 3 cm, 3 cm, 3 cm	-
2	64	M	4	Neck	3.5 cm, 4 cm, 4 cm, 4.5 cm	5 ordinary lipomas, 1 seborrheic keratosis
3	53	M	4	Neck	1.5 cm, 1.5 cm, 2 cm, 2.2 cm	-
4	47	M	1	Arm	2.5 cm	2 ordinary lipomas
5	60	M	1	Neck	1.2 cm	1 angiolipoma
6	38	M	2	Shoulder	4 cm, 3.5 cm	4 ordinary lipomas
7	64	M	2	Back	3 cm, 2 cm	2 angiolipomas, 1 FEP

A: Age, FEP: Fibroepithelial polyp, L: Location, M: Male, N: Number, P: Patient, S: Sex, SCL: Spindle cell lipoma

Clinical History

Patient 1: A 47-year-old male patient presented with 5 simultaneous painless soft tissue lesions in the right hemithorax. Five excisional biopsy materials, yellow and brownish in color and soft were sent to our laboratory. They had a maximum diameter of 2.5-3 cm.

Patient 2: A 64-year-old male patient presented with 9 simultaneous painless soft tissue lesions. Four of these lesions were at the neck and five were at the back. Four excisional biopsy materials from the neck were yellow and brownish in color, soft in consistency, the smallest measuring 3.5x3x1.5 cm and the largest measuring 4.5x3x1.5 cm. The other five soft tissue lesions from the back were homogenously yellow in color and soft in consistency, with diameters ranging between 2.5 and 5 cm. He also had a hyperpigmented skin lesion on the scalp.

Patient 3: A 53-year-old male patient presented with 4 painless soft tissue lesions in the neck. These four lesions were yellow and brownish in color, soft in consistency, and had a maximum diameter ranging between 1.5-2 cm. Clinical history revealed that his father and brother had similar soft tissue lesions.

Patient 4: A 47-year-old male patient presented with 3 soft tissue lesions in the arm. Three lesions, yellow and brownish in color, soft in consistency, with a maximum diameter ranging between 2.5-3.5 cm, were sent.

Patient 5: A 60-year-old male patient presented with 2 soft tissue lesions at the neck and lumbar region. The lesions were yellow in color and soft in consistency, with a maximum diameter of 1.5 cm each, and were sent to our laboratory.

Patient 6: A 38-year-old male patient presented with 6 soft tissue lesions at the right shoulder. The lesions, which were tan-yellow in color and soft in consistency, with a maximum diameter varying between 3 and 5 cm, were sent to our laboratory.

Patient 7: A 64-year-old male patient presented with 4 soft tissue lesions on the back. Lesions that were tan-yellow in color, soft in consistency, with a maximum diameter varying between 2 and 3 cm, were sent to our laboratory. He also had a polypoid skin lesion on his back.

An informed consent form has been received from all patients.

Pathological Findings

SCL: Five lesions from patient 1; 4 lesions from patient 2; 4 lesions from patient 3; 1 lesion from patient 4; 1 lesion from patient 5; 2 lesions from patient 6; and 2 lesions from patient 7 showed similar microscopic features, consistent with SCL. All these lesions comprised fibroblast-like bland spindle cells and mature adipocytes interspersed within a fibromyxoid stroma containing coarse collagen fibers (ropy collagen) (Figures 1 and 2). In patients 1, 2, and 5, the spindle cell component was dominant and the adipose component was minor (Figure 2). These tumors had prominent areas of myxoid degeneration. In patients 6 and 7, the tumors showed multiple myxoid degeneration areas with a maximum diameter of 2 mm (Figure 3), containing spindle cells. These tumors contained prominent adipose components. Immunohistochemically, in all tumors, the spindle cells were diffusely positive for CD34 (Figure 4). In all SCLs, except for patient 2, scattered mast cells were also observed (Figure 2). Histopathological examination revealed no atypical cells. No mitotic activity or necrosis were observed.

Additional pathological findings: In 5 patients, multiple ordinary lipomas consisting of mature fat cells displaying no atypical features were observed. Patient 2 also had seborrheic keratosis. Two patients had coexistent angiolipomas consisting of mature fat cells and narrow vascular structures. Patient 7 also had a fibroepithelial polyp.

After a maximum two years of follow-up, none of the SCL lesions showed recurrence.

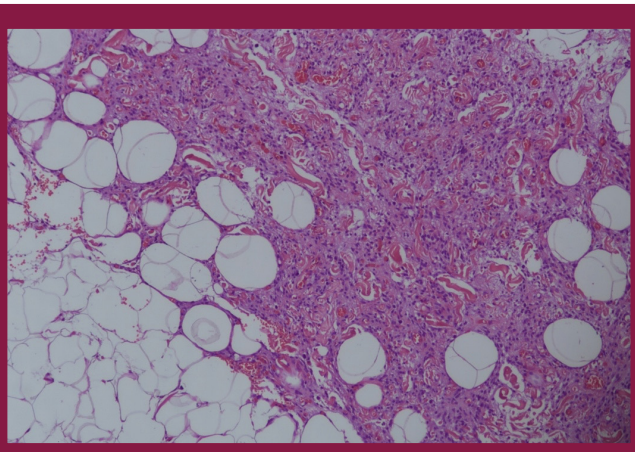

Figure 1. Spindle cells and mature adipocytes interspersed within a fibromyxoid stroma with coarse collagen fibers (x100; H-E)

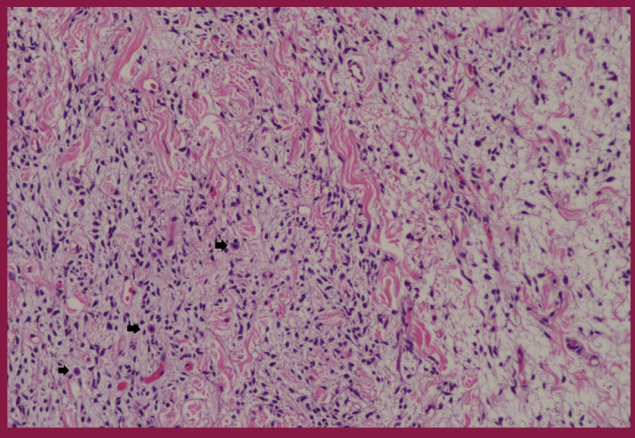

Figure 2. Prominent spindle cell component and scattered mast cells (arrows) within the fibromyxoid stroma and ropy collagen fibers (x200; H-E)

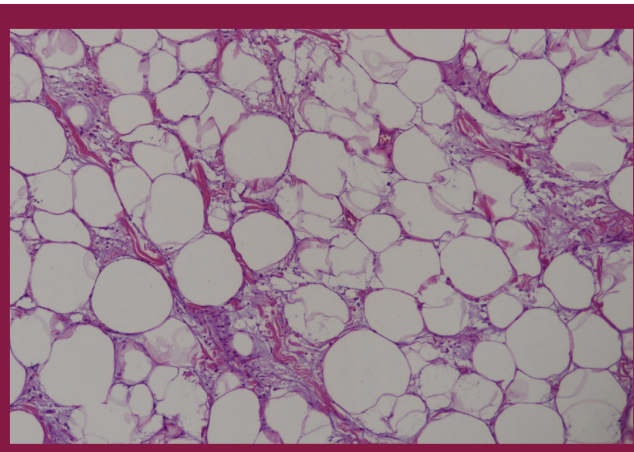

Figure 3. Small foci of spindle cell proliferation within a myxoid stroma containing coarse collagen bundles (x100; H-E)

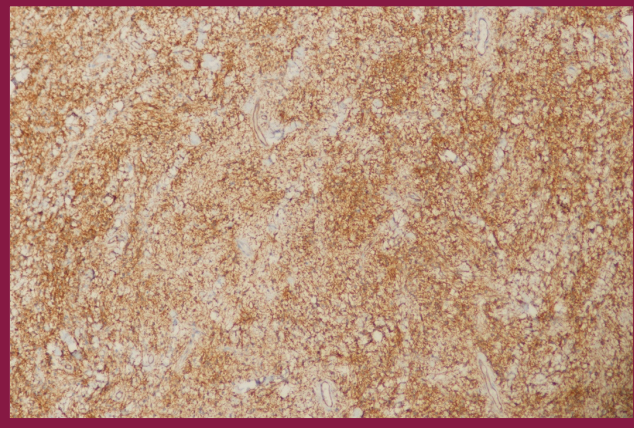

Figure 4. Diffuse CD34 immunopositivity in the SCL (100; CD34)

Discussion

SCL is a benign lipomatous tumor characteristically occurring as a solitary lesion in the posterior neck, back, and shoulders of elderly men (1). Its etiology and pathogenesis remain unclear. Multiple SCL cases are reported to be very rare. Fanburg-Smith et al. (5) reported that the ratio of patients with multiple SCLs in two different series was 0.5% and 3%, respectively. In our series, two male patients had multiple SCLs having synchronous tumors 4 and 5 in number. The distribution of lesions was limited to the trunk and posterior neck. In the English literature, multiple SCLs at the tongue and upper extremities have been reported (9,10,11). Histopathological findings were similar in both patients. It has been claimed that the natural progression of the disease may be sequential, and the later lesions may be more cellular (5). In 3 patients, concurrent lipomas and in 2 patients, coexistent angiolipomas were present. Interestingly, in two patients (patient 6 and 7), we observed multiple foci of spindle cell proliferation in a myxoid stroma. These foci had a maximum diameter of 2 mm. We believe that these areas support the suggestion that ordinary lipomas may convert into SCLs within time. Currently the mechanisms of this alteration cannot be explained. It has been reported that human mast cells activate fibroblast proliferation and collagen synthesis (12). Although SCLs in our series contained scattered mast cells except for one case, as Sakai et al. (6) reported, we also could not observe mast cells in ordinary lipomas.

Patients with synchronous multiple ordinary lipomas are reported to be approximately 5% (3). The coexistence of SCL

and ordinary lipomas (or angiolipomas) is very rare (6,7,8). To the best of our knowledge, the coexistence of SCLs and angiolipomas has not been reported before.

Histopathological differential diagnosis of SCL includes neurofibroma, dermatofibrosarcoma, angiomyofibroblastoma, superficial angiomyxoma, and myxoid liposarcoma (4,5). The diagnosis of classic SCL is usually not difficult. However, special variants and heterogeneous composition of SCLs make the diagnosis difficult (4). Diffuse CD34 immunopositivity in spindle cells is helpful in differential diagnosis (5). Fletcher et al. (13), in their report of cytogenetic analysis, found that SCLs showed chromosomal aberrations involving 16q.

Unfortunately, we could not perform any cytogenetic or molecular analysis in our cases.

In summary, we have reported multiple SCLs in 2 patients and SCL as a component of multiple lipomatosis in 5 patients. Both conditions are reported to be very rare. In 2 patients, we detected a diagnostic SCL component as multiple small foci in otherwise ordinary lipoma-like mature adipocytic cells. We believe that this finding may support the claim that ordinary lipomas may be converted into SCLs within a period of time.

Ethics

Informed Consent: An informed consent form has been received from all patients.

Authorship Contributions

Surgical and Medical Practices: C.Ç., H.B., R.T., Concept: C.Ç., H.B., R.T., Design: C.Ç., H.B., R.T., Data Collection or Processing: C.Ç., H.B., R.T., Analysis or Interpretation: C.Ç., H.B., R.T., Literature Search: C.Ç., H.B., R.T., Writing: C.Ç., H.B., R.T.

Conflict of Interest: No conflict of interest was declared by the authors.

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