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Focal Myositis as a Manifestation of Behcet's Disease: Single Center Case Series

Seung Mi Yeo[1*], Min Soo Kang[1], Duk Hyun Sung[1†]

Samsung Medical Center, Department of Physical and Rehabilitation Medicine[1]

Purpose
Behçet's disease (BD) is a systemic form of vasculitis characterized by major symptoms of oral aphthous ulcers, uveitis, skin lesions, and genital ulcers. Although BD has the capacity to exert an effect on all organs of the body, myositis is uncommon in BD and may confuse clinicians, especially if a definite diagnosis of BD cannot be made. We aimed to summarize the clinical, laboratory, histologic and image findings of patients whose initial manifestation of BD was focal myositis.

Subjects & Methods
We retrospectively investigated the clinical, laboratory, histologic and image findings of 8 patients with focal myositis associated with BD who visited the outpatient clinic of Samsung Medical Center from 2011 to 2019.

Results
The case series included 8 patients, 7 (87.5%) males and 1 (12.5%) female. The mean (±1SD) age at the time of diagnosis of the disease was 37.9 (±11.0) years (range: 21 to 55). All the patients complained of localized myalgia. Leg, especially calf, was involved most frequently (62.5%). Clinically, ocular lesion was present in 2 patients (25%), oral ulcer was present in 7 patients (87.5%), genital ulcer was present in 5 patients (62.5%), skin lesion was present in all patients (100%), neurological manifestation was present in 2 patients (25%), vascular manifestation was present in 3 patients (37.5%) and pathergy test was positive in 2 patients (25%). According to the International Criteria for Behçet's Disease, 6 patients meet the BD diagnostic criteria but 2 patients do not meet the criteria. According to the Diagnostic Criterion of the Behçet's Disease Research Commitee of Japan (2003 revised), one patient is classified as complete BD, four are incomplete BD and three are possible BD. All 8 patients in this study meet equal to or more than possible type of criteria. Laboratory tests revealed that elevated ESR/CRP in 5 patients (62.5%), weakly positive nuclear FANA in 4 patients (50%), elevated CK in 2 patients (25%), elevated ALT in 2 patients (25%) and positive HLA B51 in 2 patients (25%). Skin biopsy was conducted in 2 patients and revealed perivascular lymphocytic infiltration suitable for vasculitis. Muscle biopsy was conducted in 3 patients and only one specimen was considered reasonable for vasculitis. MRI of the

involved limb was performed in 7 patients, multifocal myositis was found in all patients (100%) and thrombophlebitis, arteritis and neuritis were also found in one patient (14.3%). Seven patients received oral prednisolone treatment with dose tapering for myositis. The starting dose of prednisolone varied from 10 mg to 60 mg, and the duration of use varied from 1 week to 1.5 years. One patient received only colchicine. All patients were pain-free after 1 to 3 weeks of short-term follow-up.

Conclusion

In the differential diagnosis of localized myositis, BD is an essential consideration. For focal myositis associated with BD, the prognosis is good because it subsides quickly to steroid treatmen

Table 1. Clinical and laboratory findings of patient with focal myositis associated with BD

	Sex/ Age	Chief complaint	International Criteria for Behçet's Disease : scoring ≥4 indicates Behçet's diagnosis								Abnormal lab finding	HLA B51	MRI	Biopsy
			Ocular lesion (2)	Oral ulcer (2)	Genital ulcer (2)	Skin lesion (1)	Neurological manifestation (1)	Vascular manifestation (1)	Positive pathergy (1)	Total (10)				
1	M/52	Myalgia of left calf	0	2	2	1	1	1 (Both L/E DVT, thrombophlebitis, arteritis)	1	8	ESR/CRP ↑ FANA nuclear weakly positive	(-)	- Thrombophlebitis with arteritis and myositis of both lower leg - Tibial neuritis	Skin biopsy - Necrosis of epidermis with ulcer - Perivascular lymphocytic infiltration in superficial and deep dermis
2	M/21	Myalgia of left calf	0	2	0	1	0	0	0	3	ESR/CRP ↑	(-)	- Myositis (high SI with enhancement) involving both lower leg	Muscle biopsy - Leukocytoclastic and necrotizing vasculitis - Lymphocytic vasculitis and perivasculitis - Multifocal interstitial infiltration of lymphocytes, eosinophils and plasma cells - Marked perimysial fibrosis
3	F/38	Myalgia of both leg	0	2	2	1	0	0	0	5	FANA nuclear weakly positive	(-)	- Myositis (high SI with enhancement) involving both lower leg	NT
4	M/38	Myalgia of left anterior thigh	0	2	0	1	0	0	0	3	ESR/CRP ↑ ALT ↑	(-)	- Myositis (high SI with enhancement) involving both lower leg	Muscle biopsy - No evidence of vasculitis is identified
5	M/41	Myalgia of left anterior thigh	0	2	2	1	0	0	0	5	ESR/CRP ↑ CK ↑	(+)	NT	NT
6	M/55	Myalgia of left calf	2	0	0	1	0	1 (Left L/E DVT)	0	4	FANA nuclear weakly positive	(-)	- Myofasitis (high SI with enhancement) involving left lower leg	Muscle biopsy - Minimal non-specific change
7	M/26	Myalgia of right calf	2	2	2	1	1	1 (Both L/E DVT)	0	9	ESR/CRP ↑	(+)	- Myofasitis (high SI with enhancement) involving right proximal lower leg	Skin biopsy - Diffuse panniculitis with multifocal vague granulomas - Erythema induratum (panniculitis, nodular vasculitis)
8	M/32	Myalgia of left upper arm	0	2	0	1	0	0	1	4	ALT ↑, CK↑ FANA nuclear weakly positive	(-)	- Myofasitis (high SI with enhancement) involving left upper arm	NT

Table 2. Treatment and prognosis of patient with focal myositis associated with BD

	Sex/ Age	Chief complaint	Treatment	Prognosis
1	M/52	Left calf pain	Azathioprine Prednisolone 60mg 1year with tapering	1week follow up – Pain free state
2	M/21	Left calf pain	Cholchicine Prednisolone 10mg 1week	1week follow up – Pain free state
3	F/38	Both whole leg pain	Cholchicine Prednisolone 20mg 4weeks with tapering	1month follow up – Pain free state
4	M/38	Left anterior thigh pain	NSAIDs Prednisolone 50mg 2weeks with tapering	2weeks follow up – Pain free state
5	M/41	Left anterior thigh pain	Cholchicine	2weeks follow up – Pain free state
6	M/55	Left lower leg pain	Prednisolone 20mg 3weeks with tapering	3weeks follow up – Pain free state
7	M/26	Right distal thigh, calf pain	Cholchicine Prednisolone 1.5years with tapering	2weeks follow up – Pain free state
8	M/32	Left upper arm pain	Cholchicine, Sulfasalazine Prednisolone 30mg 4weeks with tapering	1weeks follow up – Pain free state