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Short title: Microvenular Hemangioma

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Letter to the Editor

Dear Editor,

A 44-year-old Japanese man presented to our department for evaluation of multiple asymptomatic reddish brown macules and nodules on his face. He had been aware of them for a few years. He had no past history of acne vulgaris or injury or comorbidity. Physical examination revealed multiple red-brown flat and/or elevated macules and nodules, approximately 4 mm to 10 mm in diameter, localized on the face including the bilateral temples, preauricular areas and cheeks as well as the chin (*figure 1A*). Dermoscopy revealed a homogeneous light reddish-brown region avoiding hair follicles (*figure 1B*). We suspected that the lesions were discoid lupus erythematosus, pseudolymphoma, sarcoidosis or lichen planus. A skin biopsy was done on the left preauricular lesion (*figure 1C*). Histopathological examination revealed irregularly branched small vessels proliferating from the superficial to deep dermis. There were no atypical endothelial cells in the vessels (*figure 1D and 1E*). There were no

histopathological features suggesting rosacea and acne vulgaris. Immunohistochemically, the endothelium was positive for CD34 (*figure 1F*) and CD31 (*figure 1G*) but negative for MIB-1, podoplanin (D2-40) and GLUT-1. A final diagnosis of microvenular hemangioma (MVH) was made from these findings.

MVH is a rare acquired benign vascular neoplasm, which occurs as a solitary nodule on the extremities, particularly on the forearms or trunk.[1,2] It is reddish blue or purple to red in color.[2] Only 3% of MVH cases occur as plaques or macules,[1] which were seen in our case as well as nodules. To our knowledge, there have been only a few reports of MVH on the head and neck region (including the face).[1,3] Those cases occurred on the chin, cheek and nose.[1,3] Eleven reported cases of multiple MVHs were reported (summarized in table 1:Supplementary content). [1-4] The most preferential sites were the trunk and/or extremities (83.3%) followed by the cheek and abdomen (8.3%). Therefore, our case is a rare case of MVH exhibiting multiple tumors on the face. A previous report presented dermoscopic findings including diffuse erythema with red globules occurring on a back macule and a toe nodule.[5] Judging from those reported cases, such globules reflect the proliferation of capillary vessels. In our case, the tumor exhibited a pseudopigment network pattern composed of melanin in rete ridges and there were no recognizable globules. Our case showed atypical features of microvenular hemangioma

because of it occurrence on the face.

The histopathological features of MVH consist of proliferation of small, thin-walled, irregularly branched vessels from the reticular layer to the deep dermis. The vascular lumina are inconspicuous and endothelial cells are normal in appearance.[1,2] Our case fits these results. Clinical differential diagnosis of MVH includes neoplasm, nonvascular tumour and inflammatory lesion, including leiomyoma, dermatofibroma, lymphomatoid papulosis, lupus erythematodes (LE), morphea, papulonectoric tuberculoid, lichen planus, sarcoidosis and drug eruption.[2,3] The most important differential diagnosis is patch-stage Kaposi's sarcoma,[1,2] which is distinguished from MVH by the expression of D2-40 and human herpes virus 8 (HHV-8).[2]

We should consider the possibility of MVH in the differential diagnosis of multiple erythematous lesions.

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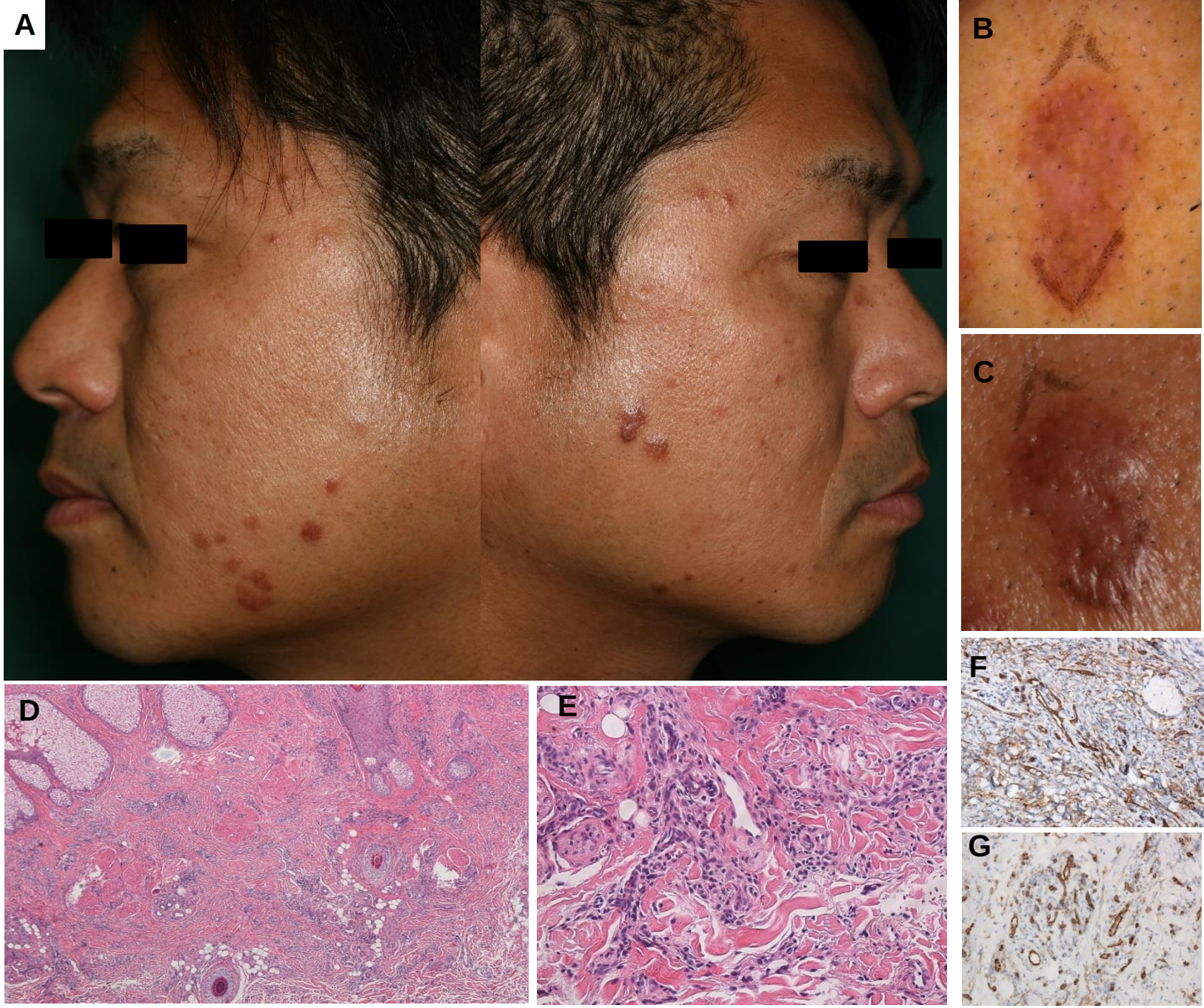
Figure Legends

Fig.1. A) Multiple reddish patches were seen over the entire face. B) Dermoscopy showed homogeneous light red-brown areas avoiding hair follicles. C) Biopsy was performed on the left preauricular lesion. D, E) Proliferation of irregularly branched vessels without atypia was seen in the whole dermis (H&E, ×40 and × 200). F, G) These cells showed positive staining for CD34 and CD31 (×200).

Table S1. *Clinical features of multiple MVHs in previously reported cases and our case*

Case (ref. No.)	Age/Sex	Location	Duration	Number	Clinical Features (Size)	Complication
1 (1)	64/F	cheek and abdomen	20mo	2	smooth, firm and reddish nodules (1.0×1.0 and 1.5×2.0 cm)	N.D.
2 (1)	55/F	chest and lower extremities	2yrs	numerous	Hemangiomas	POEMS syndrome
3 (1)	24/F	trunk	2mo	> 10	slightly tender, dark-brown papules (0.1-0.5 cm)	healthy
4 (1)	41/M	trunk and limbs	5yrs	hundreds	tender, dark-brown papules and plaque (0.1-1.0 cm)	healthy
5 (1)	38/M	neck, trunk and limbs	4yrs	40 - 60	similar to case 3	healthy
6 (1)	33/M	back	2yrs	40 - 60	similar to case 3	healthy
7 (1)	13/M	right arm, shoulder, back and chest	7yrs	multiple	erythematous nodules (up to 1 cm in diameter)	healthy
8 (1)	53/F	bilateral proximal thighs and axillae	12mo	numerous	violaceous macules, papules and plaques with vague reticulated pattern	healthy
9 (4)	35/F	trunk and limbs	5yrs	multiple (numerous) (> 100)	dark red, circular, non-scaly maculopapules (5mm in diameter)	N.D.
10(2)	43/M	left upper back and lower back	N.D.	2	separate lesions similar to prurigo nodule and atypical nevus	N.D.
11(2)	1/M	left knee	N.D.	3	agminated red-blue nodules	congenital hemihypertrophy
12(our case)	41/M	entire face (bilateral temple, preauricular area, cheek, and chin	2-3yrs	multiple	asymptomatic red-brown macules (4-10 mm in diameter)	healthy

N.D., not described, M, male; F, female.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