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Bullous pilomatricoma in young children

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Dear Editor,

Pilomatricoma is a benign tumor characterized by differentiation from hair matrix cells to hair cortical cells. It commonly occurs on the head and neck in young individuals. Although blistering is rare, when present, it is typically observed on the upper arms, shoulders, and back. The condition is more frequently seen in teenagers and young adults; however, it is rare in young children. The first case we report (Case 1) involves a 6-year-old Japanese girl who presented to our outpatient clinic with a 6-month history of a tumor on her back. On her initial visit, a physical examination revealed a 25-mm, firm subcutaneous nodule on the left shoulder, with a 20-mm, soft, dome-shaped, erythematous, broad-based pedunculated nodule directly above it (Figure 1A).

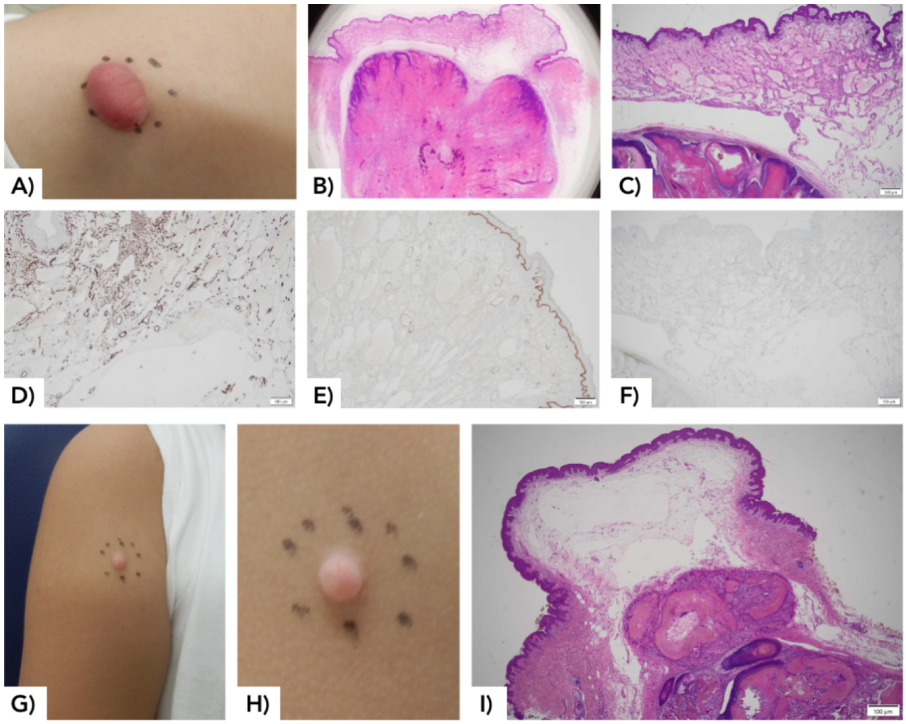


Figure 1. Clinical and histopathological findings in Case 1 (**A-F**) and Case 2 (**G-I**). **A**) A 25-mm, mobile, well-defined, firm subcutaneous nodule was located on the left shoulder extending to the upper back. A 20-mm, soft, dome-shaped, erythematous, broad-based pedunculated nodule was situated directly above it. **B, C**) The raised erythematous nodular area overlying the pilomatricoma showed a spongy appearance with a densely dilated lumen. **D-F**) Endothelial cells within the luminal-like structure were predominantly $CD34^{+}$, with partial D2-40 positivity and scattered CD31 expression. **G, H**) A 20-mm, firm, subcutaneous nodule was present on the right upper arm. A 5-mm, soft, dome-shaped, pinkish-red nodule was observed directly above it. **I**) Dilated and edematous intradermal lymphatic vessels were observed over the pilomatricoma. Hematoxylin and eosin staining is shown in panels B, C, and I, while immunohistochemical staining for CD34, D2-40, and CD31 is shown in panels D, E, and F, respectively. Original magnification: ×10 (B); ×20 (C, F, I); ×40 (D, E).

The lesion was surgically excised. Histologically, basaloid cells and ghost cells were observed in the dermis, consistent with a pilomatricoma. Additionally, extensive capillary hyperplasia was noted in the surrounding area. A densely dilated lumen surrounded by thin endothelial cells was identified overlying the pilomatricoma (Figure 1 B,C). Immunohistochemically, the endothelial cells were predominantly CD34^{+}, with some D2-40 positivity and scattered CD31 expression (Figure 1 D-F). Scattered lymphatic duct dilatations were also observed; however, the large luminal-like area directly above the tumor was negative for all markers on immunohistochemistry.

The second case (Case 2) involves a 6-year-old Chinese boy who visited our outpatient clinic with a 3-month history of a tumor on his right upper arm. On his initial visit, a physical examination revealed a 20-mm, firm subcutaneous nodule on the right upper arm, with a 5-mm, soft, dome-shaped, pinkish-red nodule directly above it (Figure 1 G,H). The lesion was surgically excised. Histologically, basaloid cells and ghost cells were observed in the dermis, consistent with a pilomatricoma. In the superficial layer, dilated and edematous lymphatic vessels were identified within the dermis (Figure 1I).

Pilomatricoma with bullous lesions is a relatively rare clinical entity, accounting for only 3-6% of all hair follicle tumors.[1] Most reported cases occur in teenagers and young adults. The most common sites of occurrence are the upper arms, shoulders, and back, with a relatively low incidence on the face, which is the usual site of pilomatricoma. Possible contributing factors for the higher prevalence in the upper arm and shoulder include frequent physical activity and falls in children, making these areas more susceptible to external forces. The histologic features of the bullous type include ductal dilatation and edema in the dermis directly above the tumor. The lumen in these cases has been reported to show variable positivity for lymphatic and vascular endothelial markers: in some cases, only lymphatic dilation is observed, while in others, no markers are expressed. Two mechanisms have been proposed to explain the blister-like appearance: (a) stenosis or obstruction of lymphatic or blood vessels due to external stimuli or tumor growth, resulting in secondary stasis and edema;[2] or (b) external forces may damage and dilate lymphatic vessels, allowing lymph fluid to accumulate in the dermis and produce a bullous appearance.[2] In Case 1, blistering was thought to result from the mechanism described in (b), while in Case 2, it appeared to be caused by the mechanism described in (a). We conducted a retrospective study of 92 patients, including the two cases presented in this report, who were diagnosed with bullous or anetodermic pilomatricoma (Table 1). Demographic and clinical data, including age, sex, and lesion location, were collected. The mean age of the patients was 21.4 years, with a median of 18 years; the oldest being 70 years,[3] and the youngest 5 years. Notably, only one case involved a child under 6 years of age.[4] No significant sex difference was observed. The most common lesion sites were the upper extremities (42.4%), followed by the trunk (38.0%), with the back (15.2%) and shoulders (14.1%) being particularly affected. No laterality preference was noted. Consistent with previous reports,[5] the bullous variant of pilomatricoma was less frequently observed on the head and neck, which are common sites for conventional pilomatricoma. Instead, it was more frequently found in regions subjected to mechanical stress, such as the shoulder and back. While most cases of pilomatricoma with bullous lesions have been documented in teenagers and young adults,[5] occurrences in young children are rare (cases of 6 years old and under, 3/92: 3.3%). These rare cases suggest that the pathogenesis of pilomatricoma with bullous lesions may vary depending on the age of onset and individual circumstances.

Table 1. Demographic data of patients with bullous pilomatricoma (N=92).

	N (%)
Sex	
Male	47 (51.1)
Female	45 (48.9)
Age (years)	
0-9	9 (11.4)
10-19	37 (46.8)
20-29	20 (25.3)
30-39	6 (7.6)
40-49	3 (3.8)
50-59	1 (1.3)
60-69	2 (2.5)
70-79	1 (1.3)
Location	
Head	4 (4.3)
Face	5 (5.4)
Neck	5 (5.4)
Trunk	35 (38.0)
Back	14 (15.2)
Shoulder	13 (14.1)
Upper extremities	39 (42.4)
Lower extremities	4 (4.3)
Laterality preference	
Shoulder	
Right	6 (54.5)
Left	5 (45.5)
Upper extremities	
Right	14 (48.3)
Left	15 (51.7)
Lower extremities	
Right	1 (50.0)
Left	1 (50.0)

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Consent for publication: the patients/patients' relatives provided written informed consent for the publication of this case report and any accompanying images.

Availability of data and materials: the datasets used and analyzed during the current study are available upon reasonable request from the corresponding author.

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