Pigmented hidroacanthoma simplex of the scalp mimicking clonal seborrhoeic keratosis

Dear Editor,

Hidroacanthoma simplex (HS) is a rare variant of eccrine poroma. Clinically, it manifests as sharply demarcated, brownish, flat or verrucous plaque, and often resembles seborrhoeic keratosis or Bowen’s disease. Histologically, it mimics clonal seborrhoeic keratosis (CSK), but can be differentiated by the density of CD1a cells inside the tumor nest. Pigmented HS is even more rare. We report a new case manifesting as a large, pigmented, seborrhoeic keratosis-like plaque on the right temporal area. Differentiation of HS from CSK was facilitated by the finding of a low density of CD1a-positive dendritic cells within the tumor nests.

A 73-year-old Taiwanese male with a history of squamous cell carcinoma on his left arm presented with a large, asymptomatic plaque on his right temporal scalp for 5 years. Examination revealed a 4.5 cm × 5 cm, well-circumscribed hyperkeratotic plaque consisting of coalescing, brownish seborrhoeic keratosis-like nodules (Figure 1A). No regional lymph node was palpable. The tentative diagnosis was seborrhoeic keratosis or Bowen’s disease. Histological examination of a biopsy specimen revealed epidermal hyperplasia with bulbous elongation of rete ridges containing large, discrete nests of uniform basosquamous cells (Figure 1B). Numerous pigmented dendrites were visible in the tumor nests (Figure 1C). The tumor cells displayed minimal nuclear atypia, however, some mitotic figures were visible. Immunohistochemical study showed that the tumor cells did not express carcinoembryonic antigen (CEA) or epithelial membrane antigen (EMA) except for occasional tumor cells showing membranous staining. The nests showed a marked reduction of CD1a+ cells, compared with the surrounding epidermis. The number of CD1a+ cells within the nests was approximately 5.42 cells/mm² (Figure 1D). For comparison, we counted CD1a cells in the nests of a typical case of CSK, which showed a much higher density of 14.50 cells/mm². The density of CD1a+ cells was lower in the nests than outside the nests. With the presence of pigmented dendrites in the tumor nests, the final diagnosis was pigmented HS. Our patient preferred less aggressive treatment, therefore, the lesion was treated with cryotherapy with satisfactory results.

HS, first described by Smith and Coburn in 1956, is an intraepidermal variant of eccrine poroma derived from the acrosyringium. HS commonly affects the elderly without sex predilection, and usually manifests as hyperkeratotic erythematous or brown plaques, most often on the lower extremities (45.7%) and the trunk (42.8%), and rarely on the head and neck (2.9%). The size of HS varies from 0.4 cm × 0.7 cm to 8 cm × 9 cm. Pigmented HS is a rare variant of HS and only six cases have been reported (Table 1). Involving the thigh, buttock, back, or knee. Interestingly, four of them were associated with porocarcinoma. HS needs to be differentiated from irritated seborrhoeic keratosis, Bowen’s disease, and basal cell carcinoma. The present case had uncommon features for HS, specifically, the lesion was pigmented, large in size, and located on the scalp, which can be easily misdiagnosed and treated as seborrhoeic keratosis.

The pathological findings of HS are characterized by the intraepidermal nests of poroid cells, also called the Borst-Jadassohn phenomenon, which is also observed in the lesions of CSK. In an immunohistochemical study of the Borst-Jadassohn phenomenon, compared with the surrounding epidermis, the number of CD1a+ cells in the tumor nests were either reduced (in CSK and Bowen’s disease), or absent (in HS). For differential diagnosis between HS and CSK, Liu et al found different densities of CD1a-positive cells in the tumor nest, low in HS (3.1 ± 1.0 cells/mm²) but much higher in CSK (19.9 ± 7.7 cells/mm²). The findings of low CD1a density in the tumor nests in the present case are consistent with the diagnosis of HS. The tumor cells in the present case did not express CEA and EMA. The tumor cells of HS sometimes express EMA, but not CEA. Perniciaro et al observed negative expression of CEA and focal expression of EMA in HS, and concluded that these markers were not helpful in identifying the tumor cells.

In a study of 70 reported cases of HS, Anzai et al reported malignant transformation occurring in seven patients (10%), mainly in the elderly (>73 years) and female patients (six of seven cases). The author found no difference in color or size between the malignant and benign lesions. Therefore, careful clinical monitoring of the lesion with additional sampling or early excision of HS has been suggested. This approach maybe particularly important in cases of pigmented HS, because most reported pigmented HS cases were associated with malignant change. In summary, we described a case of HS with several unusual features, specifically, pigmented, large in size, and located on the scalp, an uncommon location for HS. Pigmented HS can easily be misdiagnosed as seborrhoeic keratosis clinically and pathologically. It is important to recognize pigmented HS as it appears to carry an increased risk of malignant change.
Figure 1 An elderly man presenting with (A) a large, well-circumscribed plaque on the right temple characterized by coalescing brownish seborrheic keratosis-like nodules; (B) histopathology reveals many discrete, large nests of basosquamous cells in the epidermis, hematoxylin and eosin stain, ×20; (C) numerous pigmented dendrites are visible in the tumor nest; and (D) immunohistochemical study reveals only a few CD1a-positive cells in the tumor nest, ×400.

Table 1 Reported cases of pigmented hidroacanthoma simplex.

<table>
<thead>
<tr>
<th>Author (y)</th>
<th>Location</th>
<th>Size (cm)</th>
<th>Duration</th>
<th>Atypia/malignant transformation</th>
</tr>
</thead>
<tbody>
<tr>
<td>Velez et al (1994)</td>
<td>Back</td>
<td>-2</td>
<td>30 y</td>
<td>No atypia or dermal invasion</td>
</tr>
<tr>
<td>Lee et al (2000)</td>
<td>Right thigh</td>
<td>5 × 7</td>
<td>20 y</td>
<td>Malignant HS</td>
</tr>
<tr>
<td>Ueo et al (2005)</td>
<td>Left buttock</td>
<td>1.8 × 3.6</td>
<td>Decades, aggravated for 6 mo</td>
<td>Porocarcinoma</td>
</tr>
<tr>
<td>Lee et al (2006)</td>
<td>Right knee</td>
<td>1.8 × 2.0</td>
<td>Many y</td>
<td>Pigmented malignant HS</td>
</tr>
<tr>
<td>Ishida et al (2011)</td>
<td>Right thigh</td>
<td>1.7 × 1.2</td>
<td>3 y</td>
<td>Pigmented malignant HS with lymph node, liver and bone metastasis</td>
</tr>
</tbody>
</table>

HS – hidroacanthoma simplex.

References

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