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INVERTED FOLLICULAR KERATOSIS: A CASE REPORT AND REVIEW OF THE LITERATURE

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INTRODUCTION

Inverted follicular keratosis (IFK), called follicular poroma by Anglo-Saxon authors, is a rare benign adnexal tumor arising from the infundibular portion of the hair follicle^[1], It presents as a solitary nodular lesion, smooth or verrucous, and develops in photo exposed areas in elderly subjects. We report this case of Inverted follicular keratosis.

OBSERVATION

This is an 80-year-old patient with no prior history who consulted for a painless ulcerating lesion on the posterior aspect of the left forearm. This lesion had been evolving for 4 months with a rapid increase in size.

Clinical examination of the lesion found a round, firm, well-limited skin outgrowth, 6 cm in diameter with a central ulceration covered by a yellowish crust (figure 1).

Dermoscopic examination found a keratoacanthoma like pattern with keratin masses, hairpin vessels of radial arrangement surrounded by a white halo (figure 2). Skin biopsy shows an acanthosis epidermis with orthokeratotic hyperkeratosis. The underlying dermis is the site of a well-limited endophytic epithelial proliferation with the presence of swirling keratinocytes and horny plugs without nuclear atypia.

This histological appearance is suggestive of Inverted Follicular Keratosis. (figure 3 and 4).

The management consisted of a total tumor removal with placement of a flap for skin covering.



Figure 1: clinical aspect of the tumor.



Figure 2: dermoscopic aspect of the tumor.

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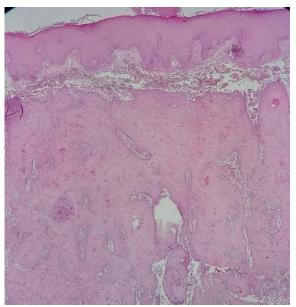


Figure 3: Endophytic Keratinocyte Proliferation Swirling

DISCUSSION

Inverted follicular keratosis was described in 1954 by Helwing. This entity was controversial for a long time because its links with irritated seborrheic keratosis, verruca vulgaris or trichilemmoma were put forward by some authors in the literature. There are exophytic forms, endophytic, flatter forms and hyperkeratotic forms. The presence of multiple KFI should lead to a discussion of Cowden's syndrome (minor criterion).

IFK occurs preferentially in adults after 60 years of age, with a male predominance. It presents as a solitary, asymptomatic, firm, well-limited, papulo-nodular or papillomato-verrucous growth with a squamous-crusted surface, occasionally with a filiform appearance like a skin horn. The clinical differential diagnosis is quite broad and includes benign tumors such as wart vulgaris, irritated seborrheic keratosis, verrucous dyskeratoma, adnexal tumors, etc., and malignant tumors such as basal cell carcinoma, keratoacanthoma, and squamous cell carcinoma, or melanoma in the rare pigmented forms. Dermatoscopic examination allows the diagnosis of IFK to be evoked, which is not done by clinical examination alone, with 3 main patterns described corresponding to the 3 histological variants. [5,6-7]

- the 1st pattern, the most frequent (60% of cases), of the keratoacanthoma or squamous cell carcinoma type, is seen in endo-exophytic or exophytic forms: it comprises a central keratin crust correlated with parakeratotic hyperkeratosis, surrounded by hairpin vessels of radial arrangement, which reflect the presence of telangiectasias within the dermal papillae, surrounded by a white perivascular halo corresponding to tumor keratinocytes and sometimes the presence of white circles.
- the 2nd pattern, solid nodule type, present in 35% of cases, is seen in the endophytic form: it comprises an area without central yellowish-white structure,



Figure 4: Higher Magnification Images Showing Keratinocytes And Horny Plugs.

homogeneous and amorphous, which corresponds to the tumor lobules, surrounded by hairpin vessels of radial arrangement surrounded by a white halo.

- the 3rd pattern, papillomatous, wart-like, is rarer and is seen in the exophytic form; and may sometimes take on the appearance of a skin horn.

Focusing on vascular structures, they are present in 100% of IFK cases, sometimes in the foreground, either as a monomorphic vascular pattern in 60% of cases or as a polymorphic vascular pattern (≥ 2 vascular structures) in 40% of cases; these may include.

- hairpin vessels of radial arrangement surrounded by a white halo, most regularly identified within KFIs, but they are also seen in other keratinizing tumors, such as seborrheic keratosis and invasive squamous cell carcinoma, some other adnexal tumors such as eccrine poroma, pilomatricoma, trichilemmoma, and even within irritated warts.
- glomerular vessels, which will be difficult to differentiate from those of squamous cell carcinoma insitu (Bowen's disease), especially in the presence of surface scales.
- -other types of vessels, rarer, more atypical: irregular linear vessels, helical vessels, globules and milky red areas.

Histologically, it is a symmetrical, well-limited lesion with ortho- and parakeratotic hyperkeratosis and sometimes a horn. [1,8] Several variants exist, correlated with the different clinico-dermatoscopic aspects: papillomatous wart-like variant (exophytic form), keratoacanthoma-like variant (exo-endophytic form) and nodular solid variant (endophytic form). IFK consists of a proliferation of large keratinocytes, distant from each other, giving a false impression of acantholysis or a "loose" appearance to the whole, arranged in lobules or digitiform projections extending to the dermis.

Dyskeratotic cells, small epidermoid coils of keratinocyte nests (onion bulbs) and small basaloid cells in the periphery are associated.

There are usually no atypia, no mitoses, no crossing of the basal area, and little or no pigment.

The histologic differential diagnosis mainly includes irritated seborrheic keratosis, trichilemmoma, and squamous cell carcinoma, in which there are more atypia and no "loose" appearance

CONCLUSION

When faced with a clinical appearance suggestive of seborrheic keratosis and a dermoscopic appearance showing a keratoacanthoma like pattern, it is wise to evoke an inverted follicular keratosis.

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