KERATOACANTHOMA WITH OSTEOLYSIS*
(A case report with an isolated interdigital lesion)

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Summary

Subungual Keratoacanthoma with osteolysis of the terminal phalanx was first described by Fisher as part of a multiple Keratoacanthoma syndrome. The case described in this paper represents an isolated interdigital lesion. Following a thorn prick a keratotic lesion evolved between the right 3rd and 4th toes of a 50 year old male. Osteolysis of the proximal and middle phalanges of the 3rd toe was demonstrated radiologically. A cup shaped lesion with buttress formation and proliferative epidermal changes in its base with dyskeratosis, para and orthohyperkeratosis and exocytosis constituted the dominant histologic findings. Partial spontaneous resolution was observed.

Keratoacanthoma a self healing pseudo-cancerous lesion was first described in its single form by Sir Jonathan Hutchinson[1]. The multiple variant was reported by Ferguson Smith[2]. Fisher's subungual Keratoacanthoma with osteolysis of the terminal phalanx[3] belongs to the latter category. The bone change has been regarded as a pressure effect of the rapidly growing tumour. Lamp et al[4] showed that healing of the skin lesion was accompanied by reversal of bone changes.

To the best of the author's knowledge an interdigital location with osteolytic changes in the proximal and middle phalanges has not been reported in the literature on keratoacanthoma.

Case report

Following a thorn prick in late 1974, a 50 year old farmer developed a boil like swelling on the right fourth toe at the injured site. The lesion rapidly evolved into a greyish white mass which spread to the web between the third and fourth toes, outer side of the third toe and to the contiguous plantar and dorsal surfaces of the foot, in the order described. The lesion stopped increasing in extent 8 months after onset. Though the lesion itself was asymptomatic, patient complained of pain in the affected toes while walking.

Examination in October 1975 one year after onset revealed a well defined greyish white plaque with overhanging edges in the web between the right third and fourth toes. The lesion extended on to the apposed surfaces of the toes and the adjoining plantar and

* Presented at the XV International Congress of Dermatology, Mexico–1977

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Received for publication on 14—2—80

dorsal aspects of the foot. Horny excrescences were present in the plaque and pressure on the overhanging edges allowed cheesy material to be squeezed from their undersurface. Both toes were swollen and scaly (Fig. 1a).

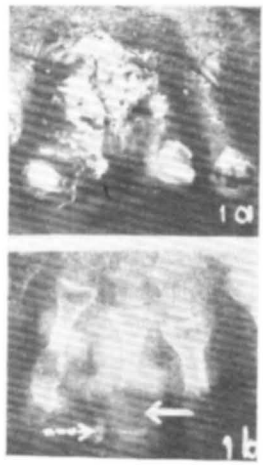

Fig. 1(a) Keratotic scaly lesion with overhanging edges and warty excrescences between the edematous right 3rd and 4th toes.

Fig. 1(b) X-Ray of the foot showing osteolysis of the proximal and middle phalanges of the 3rd toe (Solid arrow) a soft tissue shadow and areas of calcification (dotted arrow).

Roentgenograms of the foot revealed osteolysis of the proximal and middle phalanges of the right third toe. A soft tissue swelling with areas of calcification was observed around the toe (Fig. 1b).

Three punch biopsies 7 mms. in diameter were obtained from the edge of the lesion at intervals of 15 days. Formalin fixed material was studied with hematoxylin-eosin, Verhoeff-van Geison, Von Kossa, Fontana-Masson, acid orcein-Giemsa and Feulgen stains.

Sections stained with the H and E stain revealed an infolding of the epidermis upon itself, leading to the formation of a cup shaped lesion filled with keratin (Fig. 2a). The outer wall or buttress consisted of a normal to mildly hyperplastic epidermis (Fig. 2a). The dominant epidermal pathology lay in the floor and to a lesser extent in the inner wall of the cup. The basal cells and one or two layers of overlying keratinocytes showed mitotic figures. The subsequent layers of prickle cells showed poikilocytosis. Their cytoplasm was brilliantly eosinophilic and their nuclei were either pyknotic or pale staining, often lying in an unstained central portion of the cell. Spotty individual cell keratinization and formation of intraepidermal horn cysts completed the picture of disturbed keratinization. The keratinizing process began in the depth of grotesquely enlarged acanthotic rete ridges and fanned out as 'V' shaped masses into the thick stratum corneum. Adjacent rete ridges had fused in their upper part forming a common outlet for the keratinous material which was dominantly

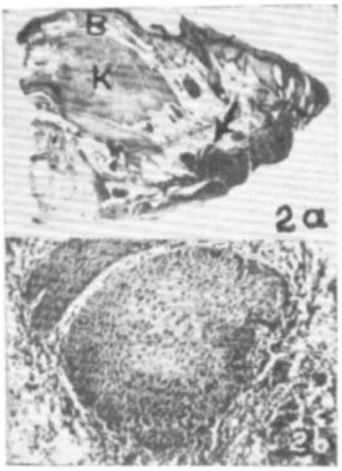

Fig. 2(a) H. E. Stain×5 shows the buttress (B) and a keratotic cup like mass (K). Arrow points to epidermal masses in the dermis.

Fig. 2(b) H. E.×100 shows well paisaded epidermal masses with a tendency to central keratinization surrounded and infiltrated by inflammatory cells.

parakeratotic. An interesting feature was the release of unevenly sized basophilic granules without formation of a granular layer (Fig. 3a). Some granules were birefringent, some gave a positive result with the Von Kossa stain, and some reacted to the Masson - Fontana stain. They were consistently Feulgen negative. With the acid orcein Giemsa stain (suggested by Dr. Hermann Pinkus) their colour varied from light blue to black. The suprapapillary epidermis, in the few areas it could be identified showed a normal granular layer with overlying orthohyperkeratosis.

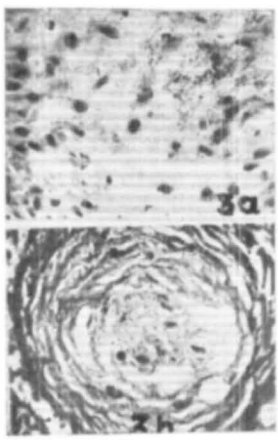

Fig. 3(a) H. E.×400 shows granules in the epidermis without a granular layer. Note dyskeratotic cells.

Fig. 3(b) H. E.×400 shows a keratinizing epidermal mass in the dermis. Note pyknotic hyper chromatic nuclei lying in vacuolated cells.

Cells from the epidermis had infiltrated into the dermis and variously sized masses were cut off from the former (Fig. 2a). They were lined by mitotically active basal cells and the central zones consisted of cells with a glassy eosinophilic cytoplasm or vacuolated cells devoid of cytoplasm and containing pyknotic nuclei (Fig. 3b).

In the region of the buttress the dermis was represented by thin streaks of connective tissue and capillaries. This and an occasional papilla were the only areas where fragmented elastic tissue and broken up connective tissue could be demonstrated with the Verhoeff-van Gieson and the Orcein and Giemsa stains. The rest of the dermis was edematous and showed infiltration with lymphocytes, plasma cells, neutrophils and large numbers of eosinophils; these being most pronounced around the epidermal masses. Lymphocytes and eosinophils were involved in the process of exocytosis in the acanthotic epidermis as well as in the epidermal masses in the dermis.

Over a period of 2 months the lesion spontaneously regressed by 30 percent and the toes became less painful. At this time the patient was lost for follow up studies.

Discussion

Several diagnostic possibilities could be considered in the case presented. It had clinical features simulating verruca vulgaris. Such a similarity was earlier noted by Montgomery while discussing Poth's paper[5]. In spite of showing clinical similarity the histology of the lesion had several features differentiating it from verruca vulgaris. Cup and buttress formation was prominent. Such a finding would be exceptional in verruca vulgaris which is a papilloma. The peripheral rete ridges of a verruca tend to converge on the centre and this was absent in the case under discussion. Keratohyaline hyperplasia a characteristic of verrucae was not observed in this patient. The dominant pathology of verruca vulgaris involves the suprapapillary epidermis leading to parakeratosis. The valleys between the elongated papillae show hypergranulosis[6]. The case under discussion showed a reversal of this phenomenon and the dominant change involved the rete ridges.

A second diagnosis to be considered was epidermoid carcinoma. The history

of rapid growth followed by a period of quiescence and final (partial) regression made this diagnosis unlikely. Bone destruction is more common with a keratoacanthoma than a slowly evolving epidermoid carcinoma. The normal arrangement of basal cells, absence of abnormal mitotic figures, exocytosis into the epidermis and normal polarity of cells tended to negate this diagnosis.

Other diagnoses which were considered and ruled out included tuberculosis, foreign body granulomas and fungal infections.

It is often included in the definition of keratoacanthoma, that the lesion originates in a pilosebaceous unit[7]. Subungual, palmoplantar and the interdigital form being recorded here, must serve as exceptions. It is usually stated that each phase of the lesion (growth, the stationary state and involution) lasts 2 to 8 weeks. However lesions of several years' duration are well documented. The case under discussion revealed them over a period of 14 months.

A history of injury prior to onset has been used as an argument to implicate an infective agent in the etiology of keratoacanthoma[7]. Though bone destruction has been attributed to a direct pressure effect of the growth[4], in the present instance edema leading to ischaemia would act as an additive factor.

Histologic changes including parakeratosis were diagnostic of a typical keratoacanthoma except that they occurred in an area normally devoid of pilosebaceous units. On the basis of staining characteristics, two types of granules viz. melanin and calcium, instead of the usual nuclear dust and keratohyaline, could be seen in the keratinizing epidermis. (Melanin is observed more often in keratoacanthoma than in epidermoid carcinomas). Vacuolated cells with pyknotic nuclei (Fig. 3b) have been regarded as the typical dyskeratotic cells of keratoacanthoma by Montgomery[8]. Repeated histologic studies from different sites of the tumour merely confirmed its benign nature.

Acknowledgements

The author is grateful to the Dean, Dr. V. N. Panse for use of hospital material, to Mr. C. V. Desai, the senior artist for the clinical pictures and photomicrographs, and Miss M. S. Shah for processing the histologic material. The author thanks Dr. Hermann Pinkus for his suggestions on identification of the granular material in the epidermis.

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