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A SWEAT GLAND TUMOUR OF THE MIXED - SALIVARY TYPE OF THE RIGHT MIDDLE TOE

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The criteria for the diagnosis of this tumour are (i) "that the tumour must arise in the skin or from sweat glands, (ii) that it must be composed of epithelial elements intermingled with cartilage or myxoid tissue in such a way that the epithelial cells appear to be continuous with these elements and not separated by a basement membrane" (Stout & Gorman 1959). Lennox et al (1952) traced the literature and found that Nasse was the first to report a case (1892), and Penisi's case (1908) was the first in which site was recorded as unmistakably in the skin. Stout and Gorman studied 39 cases and accepted another 95 cases from the world literature.

Site

Their breakdown figures regarding the sites of these 134 mixed tumours are:—

Head:	23
Face:	68
Neck:	0
Trunk:	12
Upper extremity:	12
Lower extremity:	19 (Thigh 6, Leg 6, Foot 7).

The face has the highest incidence, but a note of caution is made here so as not to include tumours of the nearby salivary, mucous or serous glands in this region. The neck, the upper arm, the anterior abdominal wall, the buttocks, the scrotum and the penis are sites where no tumour was found.

Age

It occurs at any age, but the highest incidence is between the 3rd and 5th decades.

Sex

Besides the head, the tumour is equally distributed between the sexes. In the head area it is 3:1 in favour of males.

Size

The smallest tumour was 2 x 1 mm. and the largest was 12 cm. in diameter. The latter was found on the chin of a 22-year-old Chinese who had it for 10 years.

Duration

The mean duration of the tumours was 7 years, and the extremes were 3 months and 40 years.

Malignancy

Only 1 case was accepted as a bona fide malignant tumour by these authors.

Case History of the Present Case

A 51 year old, male, Chinese, noticed a nodule on the undersurface of his right middle toe 6 years ago. It has since grown to its present size of 3″ x 2″. One month previous to the operation, a discharging sinus developed with the onset of pain. On examination it was a firm and tender mass with superficial ulceration, on to which the sinus discharged.

The right middle toe was amputated at the metatarso-phalangeal joint, and the patient had an uneventful recovery thereafter.

Gross Description

A pedunculated rounded protuberance measured approximately 5 cms. in diameter was found attached to the inferior surface of the toe. The tumour was covered by skin all round except for a localised area of ulceration approximately 15 mm. in diameter.

The cut surface was moderately firm and fleshy-looking and had a mucoid and slippery feel. It was well demarcated but not encap-

sulated. A sinus extended from the base of the ulcer to the distal phalanx through the tumour.

FIGURE I — The tumour as seen from in front.

FIGURE II — Undersurface, showing superficial ulceration and discharging sinus.

Microscopic Picture

The tumour is well circumscribed and is separated from the epidermis by a distinct layer of the dermis. In the latter are remnants of normal sweat gland structures, some of whose ducts are dilated and contain PAS — positive material.

The tumour itself is situated in the lower dermis and subcutaneous tissue, as adipose tissue becomes more abundant in the lower reaches of the tumour. The gangrenous sinus tract is lined by inflammatory necrotic tissue, and it is near this tract that the tumour is seen to approach the epidermis (Fig. III). At this junction groups of tumour epithelial cells appear to drop out from the epidermis, especially from the rete pegs (very similar to the way naevus cells drop off from the epidermis).

The tumour epithelial cells are all uniform in character. The nucleus is oval, and has well formed and mature chromatinic granules and a small basophilic nucleolus. The cytoplasm is eosinophilic and its quantity varies with the crowding and congestion of cells (i.e., it varies inversely with the number of cells). The cells are mainly spindle in shape, but in smaller aggregates they may be polygonal or cuboidal.

The tumour cells occur mostly in irregular masses, columns and sheets; in many areas concentric whorls are also seen (Fig. VIII).

The irregular masses are broken up in places by hyaline stroma, and where the epithelial cells abut stroma, they appear to transform into the different types of stromal cells, e.g., fibroblast, chondrocytes and myxomatous cells (Figs. III, IV, V, VI & VII).

It is only in rare areas that one sees tubular structures which are lined by either a single layer or a double layer of cells. This gives its sweat gland origin away.

Near the sinus tract, squamous differentiation of the tumour cells can be seen. In several loose areas oncocytes appear to be straggling in a hyaline stroma.

The stroma is mainly hyaline in type (Fig. VII). Next in frequency is the myxomatous variety (Fig. V). Collagenous, fibroblastic and

FIGURE III x 45 — Note approximation of the tumour to the epidermis near sinus tract which is just at the left edge of picture. Note droppings of epithelial elements from epidermis.

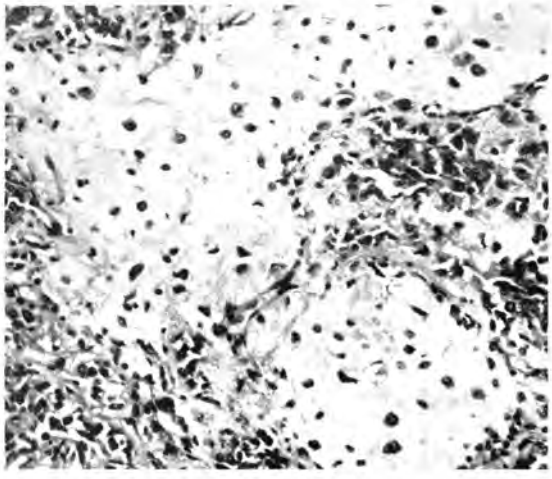

FIGURE IV x 150 — Note epithelio-cartilaginous metaplasia and incorporation of myoepithelial cells as chondrocytes.

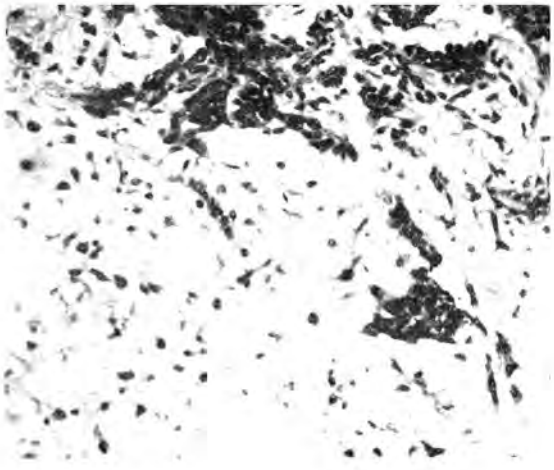

FIGURE V x 150 — Note myxomatous stroma beside groups of myoepithelial cells and incorporation of the latter into the myxoid tissue.

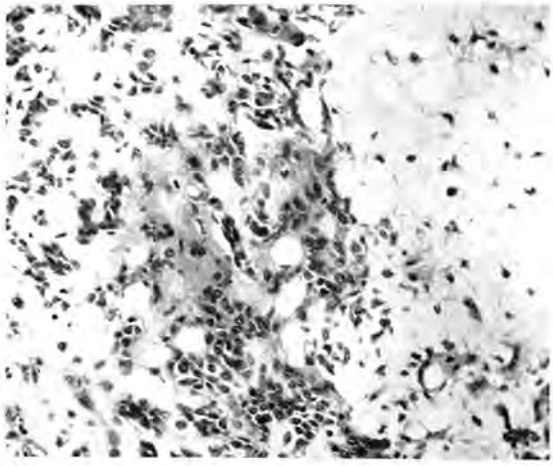

FIGURE VI x 150 — Note myxoid tissue to the right and epithelio-cartilaginous complex at left. Note also double-layered structures at bottom.

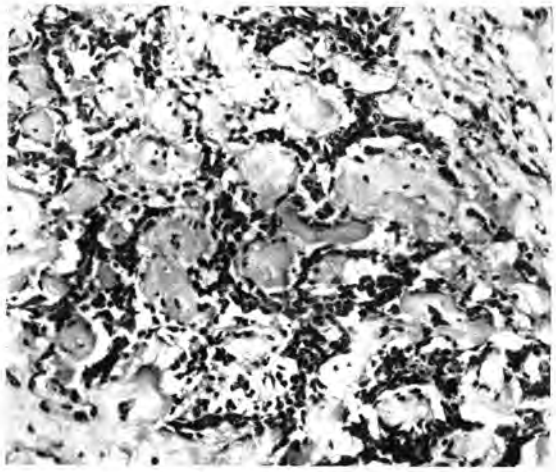

FIGURE VII x 150 — Note hyaline stroma strangling myoepithelial masses.

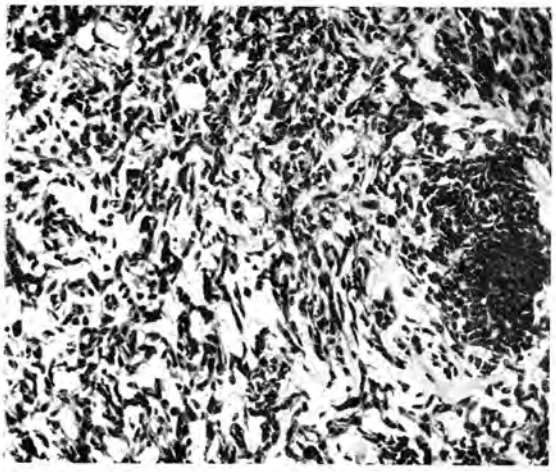

FIGURE VIII x 150 — Note typical cellular areas with a whorl of myoepithelial cells at right.

adipose varieties come next. Lastly, several groups of cartilaginous areas (Fig. IV) are also present. All the stroma cells can be seen to be fed by the epithelial cells, and in junctional areas they can be seen to be gradually included into the stroma. There is no basement membrane around any of the epithelial masses, which abut directly against the stroma, and in some places the stroma appears to be the product of the epithelial cells.

PAS and mucicarmine stains are examined and the stroma shows a much greater affinity for the PAS than the mucicarmine stains. Mucicarmine positivity appears to be greatest in the myxomatous areas and within some epithelial groups which are generating mucine. Only in these areas does one see some occasional mucicarmine positivity in some of the epithelial cells. The recently secreted substance appears most positive and remains so when not allowed to come in contact with the stroma, i.e., when they are still amidst epithelial masses. However, when in contact with the surrounding stroma, the positivity lessens and disappears completely in the well established stromal areas. The positivity also appears to concentrate around the cellular borders of the secreting epithelial cells.

Discussion

The only normal apparatus in the skin that has a double-layered structure is the sweat gland. Normally both the secretory coils and ducts are lined by an inner single layer of columnar or cuboidal secretory cells and an outer layer or layers of spindle-shaped myoepithelial cells. If in any given tumour in the skin, a double-layered structure is recognised, then one is dealing with a sweat gland tumour. The exceptions to the rule are of course when the tumour occurs in regions around the breast and salivary glands. These two organs are also blessed with the same double-layered structure.

The sole agent in producing the "mixed salivary" pattern is the myoepithelial cell (Almost all the epithelial cells in this tumour are myoepithelial cells). This myoepithelium is capable of producing a true mucin which when it comes into contact with stroma becomes a connective tissue mucin. It is by the further conversion of this so-called connective tissue mucin and the incorporation of the myoepithelial cell as the chondrocyte that true cartilage is finally elaborated.

Lennox, Pearse and Richards have followed the above changes histochemically and proposed a schematic change thus:— "epithelial mucin —— cyst mucin —— stromal mucin —— cartilage."

Thus for a tumour in the skin to be labelled as a "mixed salivary" type, it requires to have this chief criterion of epithelio-cartilaginous metaplasia.

And for such metaplasia to occur, large quantities of myxoid tissue are invariably present as well.

However, if tiny bits of cartilage are missed in the sections and there is a paucity of myxoid tissue due to small production of mucin, a tumour is most likely to be called a simple hidradenoma or a myoepithelioma. Thus a "mixed salivary" type of sweat gland tumour is no more than a hidradenoma or a myoepithelioma which has elaborated large quantities of mucin, and if given time, cartilage. In the salivary gland, pathologists are less fussy; for in this organ all myoepitheliomas are labelled as mixed salivary tumours (pleomorphic adenoma) even if there is no cartilage or large quantities of myxoid tissue!

Summary

A mixed salivary tumour of the skin of 6 years duration is described in a 51-year old Chinese.

The morphogenesis of its pleomorphic stroma is discussed.

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