Acral Syringoma: Electron Microscopic Studies on its Origin

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Abstract. A case of acral syringoma associated with the usual eruption on the lower eyelids was examined by light and electron microscopy for the first time. In the upper dermis there were a few ducts and cystic formation, while predominant atypical solid strands composed of tumor cells were often seen by light microscopy. Electron microscopy showed the formation of small ducts in the solid strands and numerous microvilli with a pinching off of the luminal tumor cells. A large number of multivesicular dense bodies (mvb) and keratohyalin granules were also found in the periluminal tumor cells. The ducts resembled embryonic intra-epidermal ducts. The unusual anatomical sites of eruption and the electron microscopic findings suggest an eccrine intra-epidermal duct origin of acral syringoma.

Key words: Eccrine intra-epidermal duct; Acral syringoma; Syringoma; Ultrastructure

Syringomas are often limited to the lower eyelids but may also involve the cheek, axilla, neck, abdomen and vulva (7). In the so-called eruptive hidradenoma the lesions are very common on the anterior trunk of young individuals (2). Reports of syringoma on the hands are rare. We found only one English report, by Hughes & Apisarnthanarax in 1977 (4) and two Japanese reports, in 1922 (10) and 1957 (5). In the former case multiple acral syringomas were limited to the dorsa of both hands. Japanese cases are associated with eruptive hidradenoma.

Recently we examined an unusual case of multiple acral syringomas associated with the usual eruption on the lower eyelids. The origin of syringoma has been disputed and its derivation from apocrine or eccrine cells is still controversial. While Hashimoto et al. (1) suggested an eccrine origin from histochemical and electron microscopical studies, Pinkus & Mehregan (8) considered an apocrine origin because of the distribution pattern which corresponded to that of the apocrine glands with sparing of the hands and feet. We have examined the fine structure of this unusual syringoma by using electron microscopy in order to contribute to a more precise determination of its origin.

REPORT OF A CASE

A 35-year-old woman was seen at the Osaka City University Medical School dermatology clinic because of a long history of acquired asymptomatic eruption on both hands and lower eyelids. Her past medical history was normal. Physical examination showed multiple, grouped, skin-coloured 2×2 mm papules on both lower eyelids and on the dorsal proximal phalanges of both hands with the exception of the thumbs. These papules formed confluent lesions and some had an annular appearance (Figs. 1, 2).

MATERIALS AND METHODS

Punch biopsy specimens obtained from the dorsal aspect of a finger and the right lower eyelid were embedded in paraffinwax and stained with hematoxylin-eosin. A part of finger specimen was fixed for electron microscopy, first with 2% osmium tetroxide in 0.1 mol/l collidine buffer at pH 7.4 for 2 hours and then post-fixed in unbuffered 6% glutaraldehyde for 2 hours. The tissue was dehydrated and embedded in Epon 812. Thin sections were stained with 2% uranyl acetate and Millonig's lead procedure and examined with the Hitachi HS-9 electron microscope.

RESULTS

The biopsy specimen from right lower eyelid showed typical findings of syringoma characterized by the presence of many solid strands of basophilic epithelial cells, while small cystic ducts were lined with two rows of epithelial cells and the ductal lumina contained amorphous debris. Some ducts possessed classical comma-like tails of epithelial cells, resembling tadpoles. However, the finger specimen showed atypical histopathologic features with round nests of epithelial tumor cells which were considered to be identical with the so-called solid strands found mainly in the upper dermis. The ductal structures were less developed than in typical cases and only very small ductal or cystic structures were observed.

Consecutive sectioning showed similar findings and typical ductal or cystic or tadpole structures were not prominent. No connection of the tumor nests with the normal eccrine sweat glands could be found but large keratinous cysts were sometimes seen (Figs. 3, 4).

Electron microscopy of the finger specimen revealed epithelial tumor cells which had numerous

tonofilaments in the cytoplasm. Small ducts were formed intracellularly and numerous short microvilli which are thought to be characteristic of sweat ducts lined their luminal walls, but no ducts were found in the intercellular spaces. The ducts were filled with secretions and contained many granules (Fig. 5). Many microvilli were released into the lumen from the luminal walls by pinching off.

In the periluminal cytoplasm, many clear vesicles (cv) and multivesicular dense bodies (mvb) were seen in that order from the luminal side, and cv formed from mvb were observed (Fig. 6). Around the existing ducts, there were often newly formed small ducts (Fig. 7). Some tumor cells contained keratohyalin droplets of varying size (Fig. 5) and numerous keratohyalin granules (Fig. 8). At some sites of the tumor cell nests, keratinization tended to be more advanced and these keratinized cells without nuclei also showed duct formation, microvilli and mvb, though the shapes of ducts were not round, but irregular.

DISCUSSION

Syringoma generally occurs in the lower eyelids of females and is also often found on the cheek, axilla, abdomen and vulva. Eruptive hidradenoma which shows widespread occurrence of syringomas mainly on the front of the trunk and in some cases, arms, thighs, back, etc. are included. There are few reports of syringoma on the dorsa of the fingers. Hughes & Apisarnthanarax reported only one case in English (4). In Japan, Tanimura (10) and Kurihara et al. (5) reported syringoma on the dorsa of the fingers associated with eruptive hidradenoma, but they had no histological confirmation. Our case is unique in that the syringomas of the dorsa of the fingers were associated with the usual eruption on the lower eyelids and the finger eruption showed an annular arrangement. Hughes et al. (4) reported no obvious differences between the common syringoma and acral syringoma. In our case, some small differences were found, consisting of a few ducts or minor cyst formation and a predominance of rounded solid strands. The poor duct formation could be due to immature differentiation with an inability to form large ducts. Further cases must be examined to decide whether these findings are characteristic for acral syringoma.

An apocrine or eccrine origin is still undecided. Pinkus et al. suggested an apocrine origin because syringoma occurs predominantly in orbital skin, an area in which well-developed Moll's glands and rudimentary apocrine glands occur (8). If the tumors are widespread, there is a predilection for the apocrine triangle between the axillae and pubes, including the vulva. They also emphasized that while the tumors may spread laterally to the flanks, their occurrence on the back of the flank and the distal extremities are rare. Including our case and other Japanese cases, four syringomas occurring on the fingers have been reported. By careful physical examination, further cases of acral syringoma may be found. These facts do not support the apocrine origin of the tumor suggested from the sites of occurrence. Using histochemical and electron microscopical methods, Hashimoto et al. insisted on the eccrine origin of the usual syringoma and eruptive hidradenoma (1). They emphasized the similarity between syringoma and embryonic eccrine intraepidermal ducts. The electron microscopical evidence for this is based on 1) the formation of intracellular duct, 2) large number of mvb, 3) numerous short microvilli that pinch off, 4) abundant periliminal filamentous bands, and 5) the presence of keratohyalin granules and keratinous cells.

However, more recent reports of morphological studies on apocrine ducts have challenged this view. In the apocrine intrafollicular duct which corresponds to the eccrine intraepidermal ducts, it has been found that 1) there is intracellular duct formation, 2) there are a few mvb (3), and 3) there are keratohyalin granules and keratinous cells (6, 9). These new findings suggest that the microscopic criteria are relative and not absolute. The electron microscopical investigation of the acral syringoma which we report here revealed a similarity to the structure of embryonic intra-epidermal ducts in two respects: 1) the presence of many mvb, and 2) the numerous microvilli which pinched off.

The unusual anatomical location of the tumors and the electron microscopical findings suggest an eccrine origin of syringoma, especially acral syringoma.

ACKNOWLEDGEMENT

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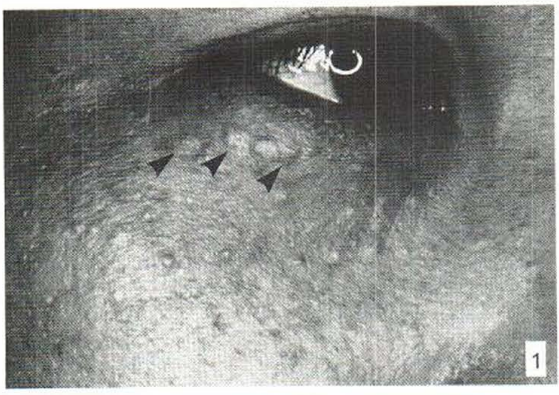

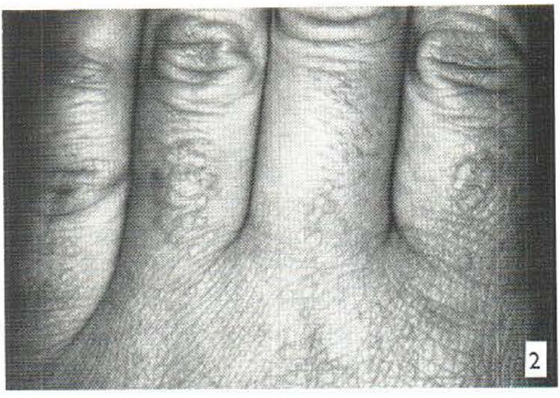

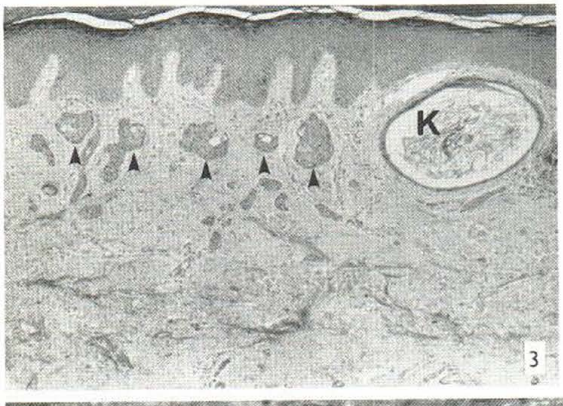
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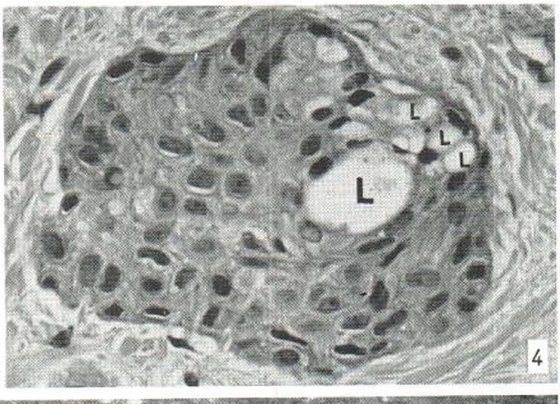
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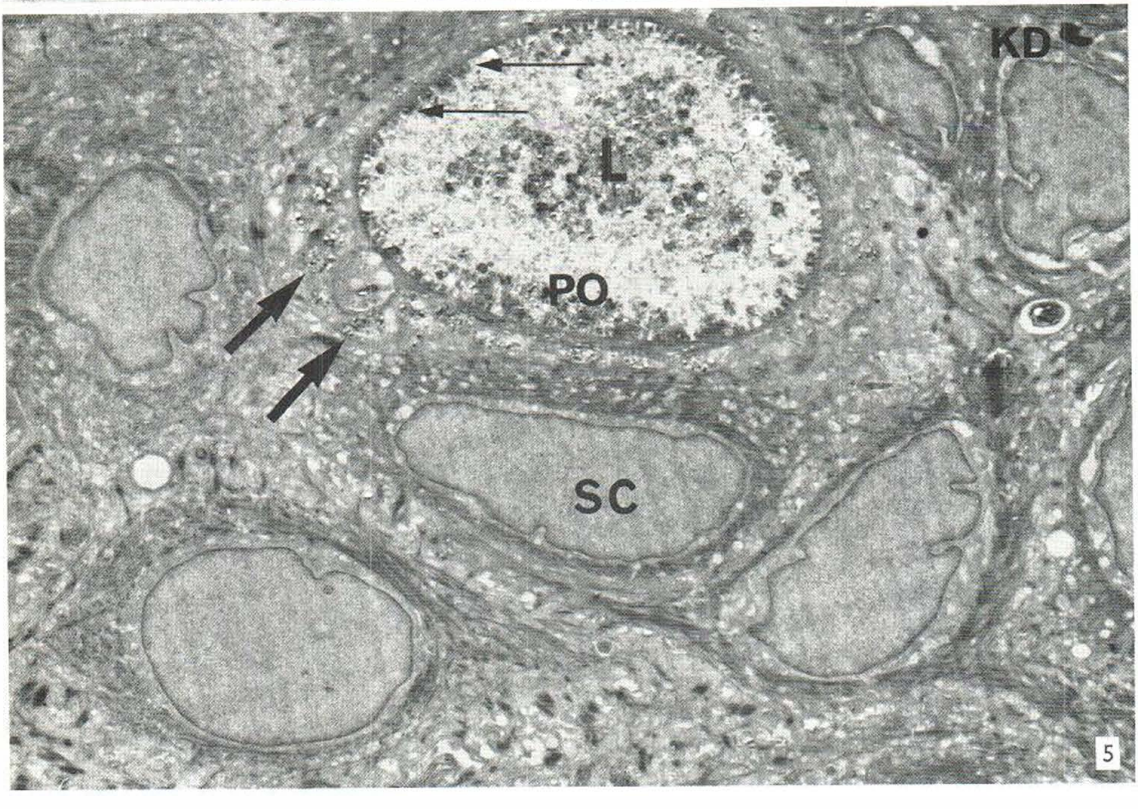
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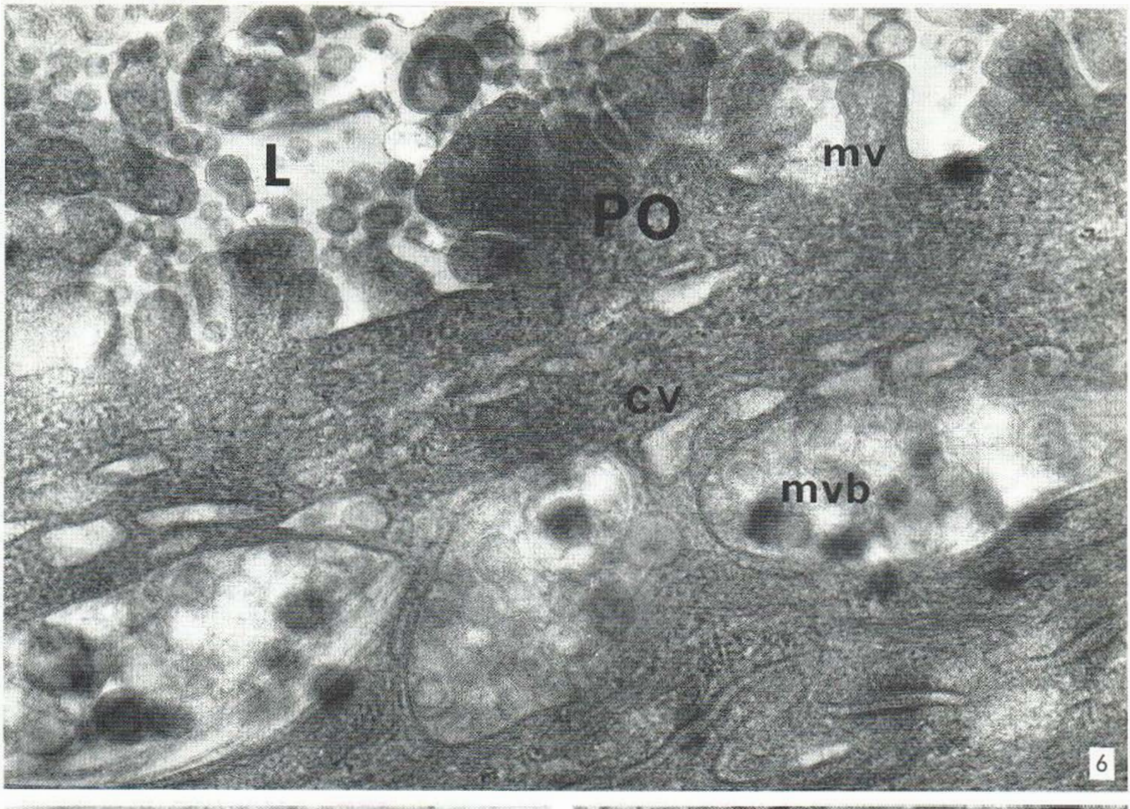

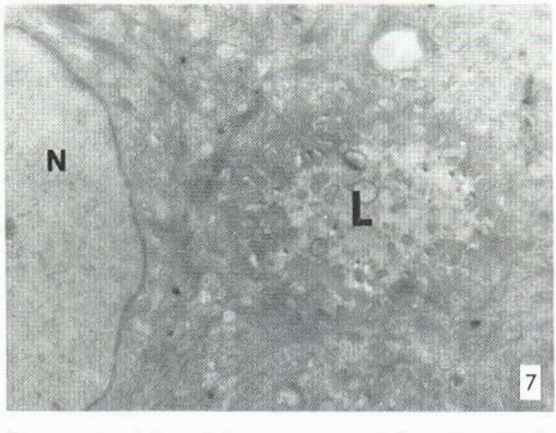

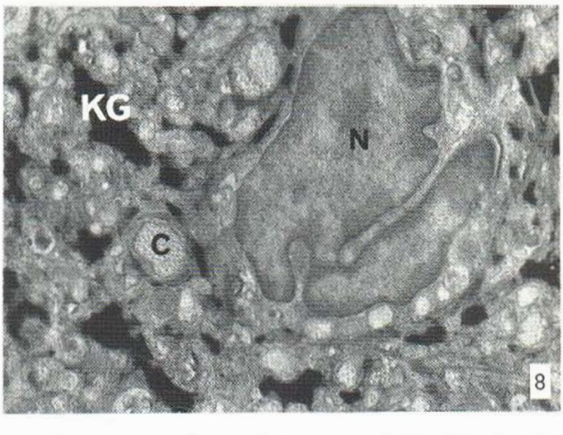

Fig. 1. Typical syringomas on the right lower eyelid (*arrowheads*).

Fig. 2. Multiple, annularly arranged, confluent syringomas on the dorsal phalanges.

Fig. 3. Histopathologic features of a papule of the right little finger, showing several atypical round solid strand (*arrowheads*) in the upper dermis (×40).

Fig. 4. Higher magnification of the strand shown in Fig. 3, showing small ductal lumens (*L*) in the tumor nest (×400).

Fig. 5. Electron micrograph of syringoma cells (*SC*), showing numerous microvilli (*thin arrows*) and their pinching off (*PO*) in the ductal lumen (*L*). Many multivesicular dense bodies (*thick arrows*) are seen in the peri luminal wall and a keratohyalin droplet (*KD*) is shown (*upper right*) (×4 700).

Fig. 6. Higher magnification of peri-luminal multivesicular dense bodies (*mvb*) shown in Fig. 5, showing clear vesicles (*cv*) formed from mvb. Microvilli (*mv*) and their pinching off in the luminal wall are also seen (×68 000).

Fig. 7. Initial small duct formation (*L*) is seen in the intracellular cytoplasm. *N*, nucleus (×9 500).

Fig. 8. Showing a syringoma cell containing many keratohyalin granules (*KG*). *N*, nucleus; *C*, collagen (×6 500).

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Banded Structure in Solitary Trichoepithelioma

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Abstract. Electron microscopical observations were made in a case of solitary trichoepithelioma. Enlarged ERs were found in the tumor cells, showing a peculiar structure with fine granules and an electron-dense banded structure, about 50 nm wide respectively, lying at intervals of 250 nm. In addition to this structure, dense ovoid bodies, about 500 to 600 nm in diameter, were also observed.

Key words: Solitary trichoepithelioma; Enlarged ER

Solitary trichoepithelioma is a trichogenic hamartoma seen predominantly on the face. This condition occurs more rarely than trichoepithelioma papulosum multiplex. Ultrastructural observations of trichoepithelioma have been rare (4, 6).

The purpose of this paper is to describe a peculiar structure; enlarged ERs found in the tumor cells of solitary trichoepithelioma. This structure resembled that described by Suzuki (7) in trichoepithelioma papulosum multiplex. We will discuss whether the structure described by Suzuki and the one we found were identical or not and whether or not these structures are specific in this condition.

CASE REPORT

A 45-year-old Japanese woman visited the Department of Dermatology, Kumamoto University Hospital in February 1978, because of an asymptomatic nodule on the right ala nasi of 4 to 5 years' duration. After long remaining rice-corn in size, the nodule had then grown for the past one year. Examination revealed an elastic, hard, pink-colored nodule, 8 mm in diameter and 6 mm in height, with teleangiectasia. On the top of the nodule, several yellowish points were noted. There was neither umbilication, ulceration, nor were hairs attached to the lesion (Fig. 1).

MATERIALS AND METHODS

The nodule was removed under local anesthesia and was cut into two pieces. One was fixed in 10% neutral formalin for light microscopy, while the other was cut into 1 mm cubes, fixed in 2% osmium tetroxide buffered solution for 2 hours for electron microscopic observation, dehydrated through a series of graded ethanol solutions, and then embedded in Epon 812.

Ultrathin sections, made on a Porter-Blum MT 2, were double stained with uranyl acetate and lead citrate. Electron microscopical observations were made with a Hitachi 12 A electron microscope.

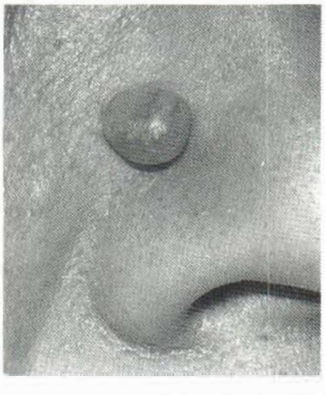

Fig. 1. Clinical picture of a solitary trichoepithelioma in a 45-year-old woman. A pink-colored elastic hard papule, 8 mm wide and 6 mm high, is present, with teleangiectasia and several yellowish points.