

PILO-SEBACEOUS CYSTIC ECTOPY OF THE UTERINE CERVIX

G. FICHERA - A. SANTANOCITO

Institute of Pathology - University of Catania (Italy)
Chair of Anatomic and Histologic Pathology IV

Summary: The infrequent finding of pilo-sebaceous cysts in an uterine cervix is presented. The different histopathogenetic hypothesis and Literature reports are critically reviewed.

The histogenesis can be referred to ectodermic embryonal germs which come in the ectopic site following an abnormal cephalic migration.

Also stressed is the possibility that neoplasms of the pilo-sebaceous structures could arise in the ectopic site and, thereafter, due to their rarity, might be under-diagnosed at the histology.

Key words: Pilo-sebaceous cyst in the uterine cervix. Uterine cervix and ectodermic structures.

The finding in a specimen of a structure belonging to the pilo-sebaceous complex in the uterine cervix is a rare event. Similarly the presence, in the same site, of pilary or glandular sebaceous structures as isolated entities is quite exceptional.

The lesion is not usually related to pathological situations of particular relevance. However, it deserves to be pointed out not only as a mere anatomical rarity or because it leads to a critical embryogenetic analysis of this area, but also because benign or at times slightly malignant neoplasias of the pilo-sebaceous structures, typical of the skin, might originate from it.

These tumors, in this specific site, might remain undiagnosed or erroneously considered as belonging to much more malignant classes of neoplasia.

EMBRYOLOGICAL NOTES

As we know, in embryos at the sixth week (18 mm) Müllerian paramesonephric ducts are formed laterally to the Wolffian mesonephric ducts by an invagination, at the cranial extremity, of the coelomic epithelium.

These, in the following phase of embryonal development migrate to a caudal direction, and running laterally to the mesonephric ducts, cross them ventrally and then lie quite medially to them.

In embryos at the eighth week (31 mm) the paramesonephric ducts, joining caudally along the medial line, form the utero-vaginal canal which, in the terminal

part, lays on the posterior surface of the urogenital sinus of cloacal derivation, thus forming the Müllerian tubercle. This latter and the urogenital sinus, once they join, form respectively the proximal four-fifths (mesodermic) and the distal fifth (entodermic) of the vagina.

The urogenital sinus, derived from the cloaca, touches ventrally the ectoderm and forms the cloacal membrane.

At this point the perineal spur, reaching the cloacal membrane, divides it into the urogenital membrane frontally and into the anal membrane behind. At the same time the cloaca becomes divided into the anterior (urogenital sinus) and posterior (terminal part of the intestine) parts.

INDIVIDUAL CASES

B. N. A 42766. Clara C., aged 42.

There is the history of a full term ectopic pregnancy, one miscarriage at the 3rd month, and uterine leiomyomatosis.

Because of protracted metrorrhagia, related to the leiomyomatosis, and having observed no improvement with medical treatment, a hysterectomy was performed. Here we refer only to the cervical specimen.

Chronic cystic cervicitis, with some areas of hyperplasia of reserve cells, was shown microscopically.

In the context of the portio we found, besides some small Naboth cysts, three round like cystic formations, 3, 8 and 12 mm of diameter, with yellowish content, poltaceous and oily to the touch and with a characteristically slightly rancid odor.

The cystic formations appeared coated with flat stratified epithelium, almost without intercellular bridging.

The cells of the wall were sometimes missing in the stratum granulosum and towards the lumen cells were large, swollen and pale in color. In some areas, while maintaining a central position, the nuclei usually became small and hyperchromatic, finally disappearing altogether, while large, clear and rounded vacuoles appeared in the cytoplasm. Their presence, even if at some points recalls sebaceous maturation, in other was similar to those found in pilar cysts (sebaceous cysts) commonly encountered on the epidermis.

The cystic content was amorphous, eosinophilic or even basophilic, sometimes arranged in irregular masses, and at some points presented needle-like accumulations of fatty acid crystals and minute calcified deposits.

Immunohistochemical tests with anticytokeratin antibodies gave positive results in the intermediate strata of epithelial lining. This positivity was reduced in the more superficial layers. Tests were negative on cystic content.

In one section, we observed a cystic formation which, by means of a short duct whose lining showed the early aspects of sebaceous maturation, opened onto the cervical cervix, coated by the normal flat, stratified epithelium.

The structures involved seemed, in fact, to derive from a limited part of the pilo-sebaceous structure, particularly from the lower part of the follicle, reaching far enough to include the opening of the sebaceous duct.

We, therefore, consider that what we have described may come within the "catagen" cysts, whose coating resembles the basal portion of the pilar follicle: the remaining aspects are similar to those of the so-called sebaceous cysts.

DISCUSSION

The presence in the ectopic site of these cutaneous adnexa has been described in Literature, although infrequently.

It is worth remembering that the sebaceous glands of the mucocutaneous junctions lie nearer to the surface than those associated with pilar structures and their

excretory duct has an isolated opening on the surface, as does in the lips, in the internal fold of the prepuce or in the labium minus.

Watson and Coll. ⁽¹⁾ described the case of a pluripara, aged 50, who wore a prolapse pessary and who had previously had a diagnostic curettage, in which the lesion of the portio was associated with the presence of ectopic sebaceous glands in the mucosa of lips and mouth. Into the portio these glands were located below the squamous lining and co-existed with a chronic cervicitis.

The AA. pointed out that in their case (and in the majority of the cases found in Literature) the presence of the sebaceous gland is not associated with the presence of mature or rudimental pilar follicles or of sweat glands, and they infer a metaplastic origin secondary to a protracted flogistic stimulation.

Chiarelli and Coll. ⁽²⁾ recently observed pilo-sebaceous structures of macroscopic pseudo-polypoid appearance in the uterine cervix of a 45 year-old woman who had not previously undergone instrumental intervention. These AA. note that the average age of incidence is between 34 and 55 years, and sebaceous glands have been described both in the exo- and endocervices.

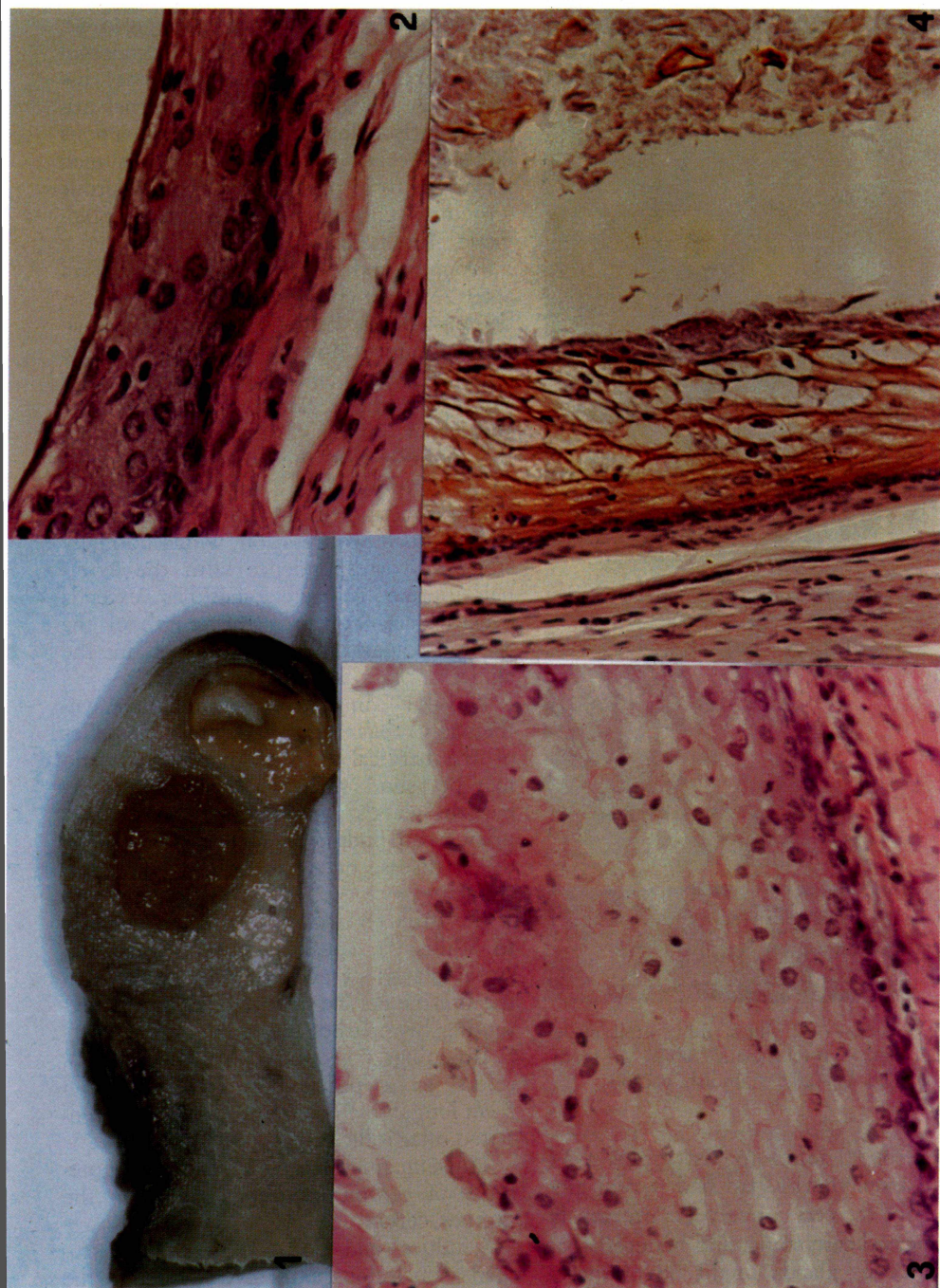
Although taking consideration of the possibility of a histogenic origin from an anomalous squamous metaplasia in the course of chronic phlogosis, they believe that instead of the metaplastic hypothesis, the origin from casual inclusions of ectodermic embryonal germs is more acceptable.

Fig. 1. — Neck of the uterus. Two large cystic formations of poltaceous content are evident.

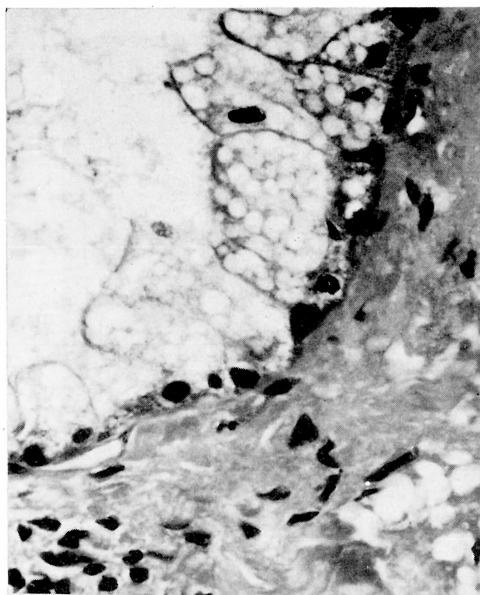
Fig. 2. — In some points, the covering consists of squamous-type cells, with few granulous elements. Tendency to surface maturation is slight.

Fig. 3. — The covering of the cysts in some points appears to consist of full, swollen, clear cells with hardly noticeable intercellular bridging.

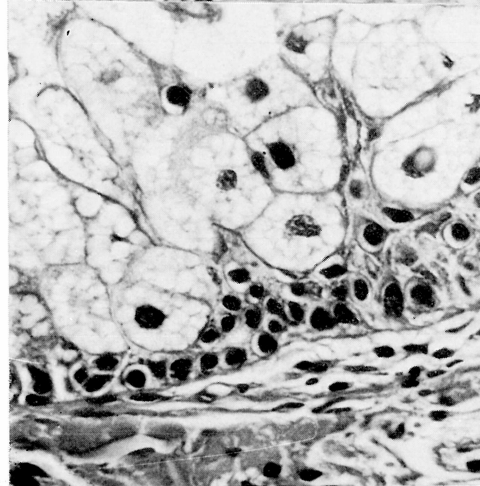
Fig. 4. — The test with antikeratin antibodies is negative in the cystic content, which is amorphous, sometimes with small irregular masses, with needle-like crystals of fatty acid.



5



6



Figg.s 5 and 6. — At times, the central nuclei are small and hyperchromatic while the cytoplasm is filled by large roundish vacuoli.

Nicholson⁽³⁾ refers to his case of a 34 year-old woman who had had partial amputation of the cervix five years previously for suspected carcinoma, and believed that the sebaceous glands might be of mesodermic derivation.

Dougherty⁽⁴⁾ found a single sebaceous gland in the cervix of a pluripara, aged 42, with uterine prolapse, who had undergone a cervical cauterisation six years before. The gland was at the external orifice slightly below the stratified squamous epithelial coating, and the Author assumed its origin from mesoblastic or entodermic structures, concomitant with chronic phlogistic stimulation.

Donnelly and Coll.⁽⁵⁾, in a pluripara aged 55, with chronic cervicitis, found sebaceous glands below the stratified squamous lining, following a bioptic sample obtained with conization. The AA. consider that these glands derive from the entodermic lining of the cervix, in that they believe that the squamous lining is to be considered as a migration in a cephalic direction of the lining of the fetal cloaca. Such proliferation, which normally stops in correspondence with the external uterine orifice, sometimes extends even up to two-thirds the length of the cervical canal, and may persist in the adult.

Novak⁽⁶⁾, in his treatise, mentions synthetically that rare segmental anomalies of the cervical epithelium of the portio may be found, such as the presence of sebaceous glands, which should not be surprising, on finding in such a site a Fordyce's disease or a basal cell adenoid carcinoma.

De Brux⁽⁷⁾ too, referring particularly to Willis⁽⁸⁾, speaks of sebaceous and sweat glands or even of pilar follicles in the uterine cervix, without, however, presenting histopathogenetic hypotheses.

Bruenings⁽⁹⁾, quoted by Nicolson⁽³⁾, refers to a polypoid cervical formation 3 cm long, which contained a well-differentiated pilar follicle.

Hinselmann⁽¹⁰⁾ too, describes the presence of ectopic sebaceous glands in the uterine cervix.

Giuducci and Coll.⁽¹¹⁾, who have studied such lesions extensively, refer to the localisation of ectopic sebaceous gland

structures in the buccal mucosa, in parotid gland, orbit and in both male and female genitals.

Fordyce⁽¹²⁾ has given the classical description of sebaceous glandular ectopy in the labial and buccal sites.

Whitehead⁽¹³⁾ refers to Salgado and Coll.⁽¹⁴⁾ in quoting the rare presence of sebaceous glands in the esophagus, attributing it to erroneous development.

De la Paya and Coll.⁽¹⁵⁾ report four cases in which sebaceous glands were in the esophageal mucosa, while Trodah and Coll.⁽¹⁶⁾ have described it in the tongue.

CONCLUSIONS

The mesonephric ducts in the female sex persist, even if residually, after birth. They appear in the form of small epithelial islets, of short tubules or small cysts, localised beyond the ovarian hilus even in the walls of the lower segment of the uterus, in the cervix and within the lateral walls of the vagina.

However, the fact that the ectopic structures we have described were always located immediately below the squamous lining (sometimes with a clearly recognisable excretory duct which opened onto the surface and showed good functional maturity), has led us to exclude a mesonephric derivation for these sebaceous structures inasmuch as in these cases the site would have been within the walls.

The hypothesis of a secondary implantation, following cervical laceration due to delivery or surgery, does not seem acceptable, since this type of history was rarely found. In other cases in which no cervical lacerations were reported, no significant histological modifications of the structure were found, excluding those due to chronic phlogosis.

Besides, in one case, a particularly accurate examination showed the contemporary presence of sebaceous ectasy in labial and buccal site.

Therefore, not all AA. agree that only

the distal part of the vagina derives from the uro-genital sinus, since some of its cells might have moved higher up, on account of focal anomalies of cephalic migration.

The discovery of sebaceous glands (or cysts) and pilo-sebaceous structures below the lining, at times with the excretory duct that opens onto the surface, leads us to hypothesize their origin from ectodermic embryonal germs.

We considered it is probable that at the time when the perineal spur subdivides the urogenital membrane forming in its upper part the urogenital sinus, ectodermic embryonal germs might have reached the ectopic site through an anomalous upward movement of part of the cloacal membrane, in whose external (caudal) face there might have adhered some of these elements.

BIBLIOGRAPHY

- 1) Watson A., Cochran A.: *J. Path.*, 98, 87, 1969.
- 2) Chiarelli S., Onnis G. L.: *Clin. Exp. Obst. Gyn.*, 8, 15, 1981.
- 3) Nicholson G.: *J. Path. Bact.*, 22, 252, 1918.
- 4) Dougherty C.: *J. Path. Bact.*, 60, 511, 1948.
- 5) Donnelly G., Navidi S.: *J. Path. Bact.*, 62, 453, 1950.
- 6) Novak E., Woodruff D.: "Gynecologic and Obstetric Pathology". Edit. Saunders C., Philadelphia, 1958.
- 7) De Brux J.: "Istopatologia ginecologica". Edit. Masson Italia, Milano, 1986.
- 8) Willis R.: "The bordeland of embryology and pathology". Edit. Butterworth Co., London, 1958.
- 9) Bruenings: *Zentl. Gynak.*, 23, 1278, 1899.
- 10) Hinselmann H.: *Zentbl. Gynak.*, 63, 1447, 1939.
- 11) Giuducci A., Hyman A.: *Dermatologica*, 125, 44, 1962.
- 12) Fordyce J.: *J. Cutan. Genito-urin. Dis.*, 14, 413, 1896.
- 13) Whitehead R.: "Mucosal biopsy of the Gastrointestinal Tract". Edit. Saunders C., Philadelphia, 1985.
- 14) Salgado G., Andrade Filho J. *et al.*: *Gastrointestinal endoscopy*, 26, 150, 1980.
- 15) De la Paya S., Pickren J.: *Archs. Path.*, 73, 397, 1962.
- 16) Trodah J., Albiorg L. *et al.*: *Archs. Derm.*, 95, 387, 1967.