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Case Reports

Hidrocystoma and Adenoma of Apocrine Anal Glands*

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Summary. Five cases of apocrine hidrocystoma of the anal glands are described, two of which are dealt with in detail, as they were a primary cause of the patients' complaints. The three additional cases were found incidentally during histologic examination of hemorrhoidectomy specimens. All five patients were women between 32 and 65 years of age. The similarities with the better known cutanous hidrocystomas of other sites harbouring apocrine sweat glands are emphasized and the morphogenesis of these lesions is considered. In addition, a rare adenoma of the apocrine anal glands in a 46 year old woman is described and the problems of diagnostic classification in examples lacking predominantly papillary structures are discussed.

Key words: Apocrine anal glands – Hidrocystoma – Adenoma – Papillary – Tubular.

Introduction

Apocrine glands seem to produce a secretion by decapitation of the apical cytoplasmic portion. The secretory product, acted upon by bacteria, may have the purpose of a pheromone. On the human body, these glands are found in the axillae, areolae, around the umbilicus, on the head, and in the anogenital region. Tumors of this type of cutaneous adnexal glands are rare, giving rise to a confusing terminology, and may cause problems in diagnosis and classification.

Case Histories

1. A 41-year-old woman reported difficulty during defecation. By proctoscopy a small prominence was seen at the ano-rectal mucosal border. This was diagnosed clinically as a polyp and removed with a biopsy forceps.

* Dedicated to Prof. Dr. Georg Klingmüller on the occasion of his 60th birthday

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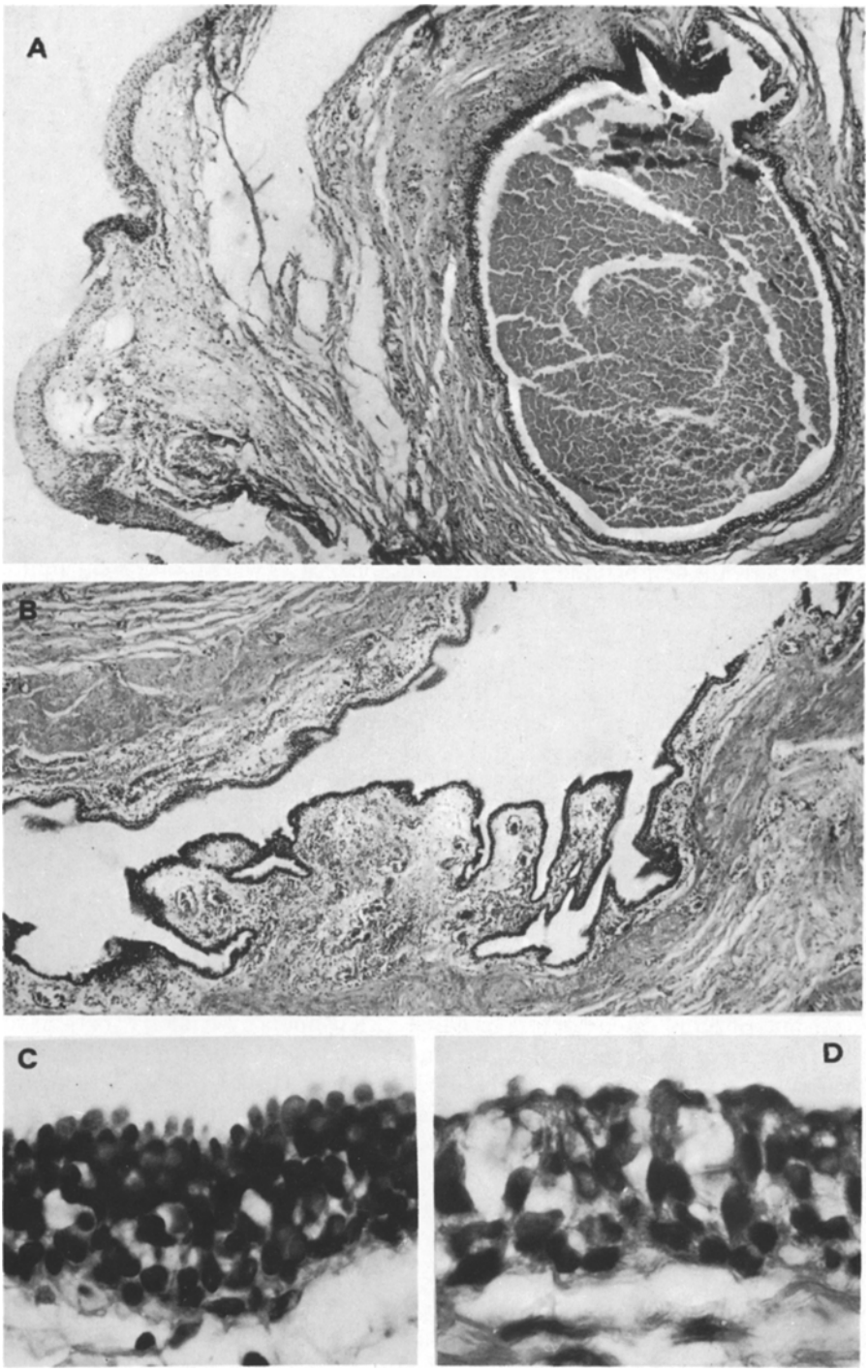

Fig. 1a–d. Apocrine hidrocystomas of anal glands. **a** shows a cystic space in the stroma underlying anal mucosa containing PAS-positive secretion and lined by an actively secreting epithelium with some clearly recognizable "hob-nail" cells characteristic of apocrine decapitation secretion (**d**). **b** shows a part of the deeper hidrocystoma of our case 2 with rudimentary papillary formations and a typical apocrine epithelial content (**c**). **a** and **b** H & E., ca. ×50; **c** PAS-Hematox, ca. ×640; **d** PAS-Hematox, ca. ×1,025

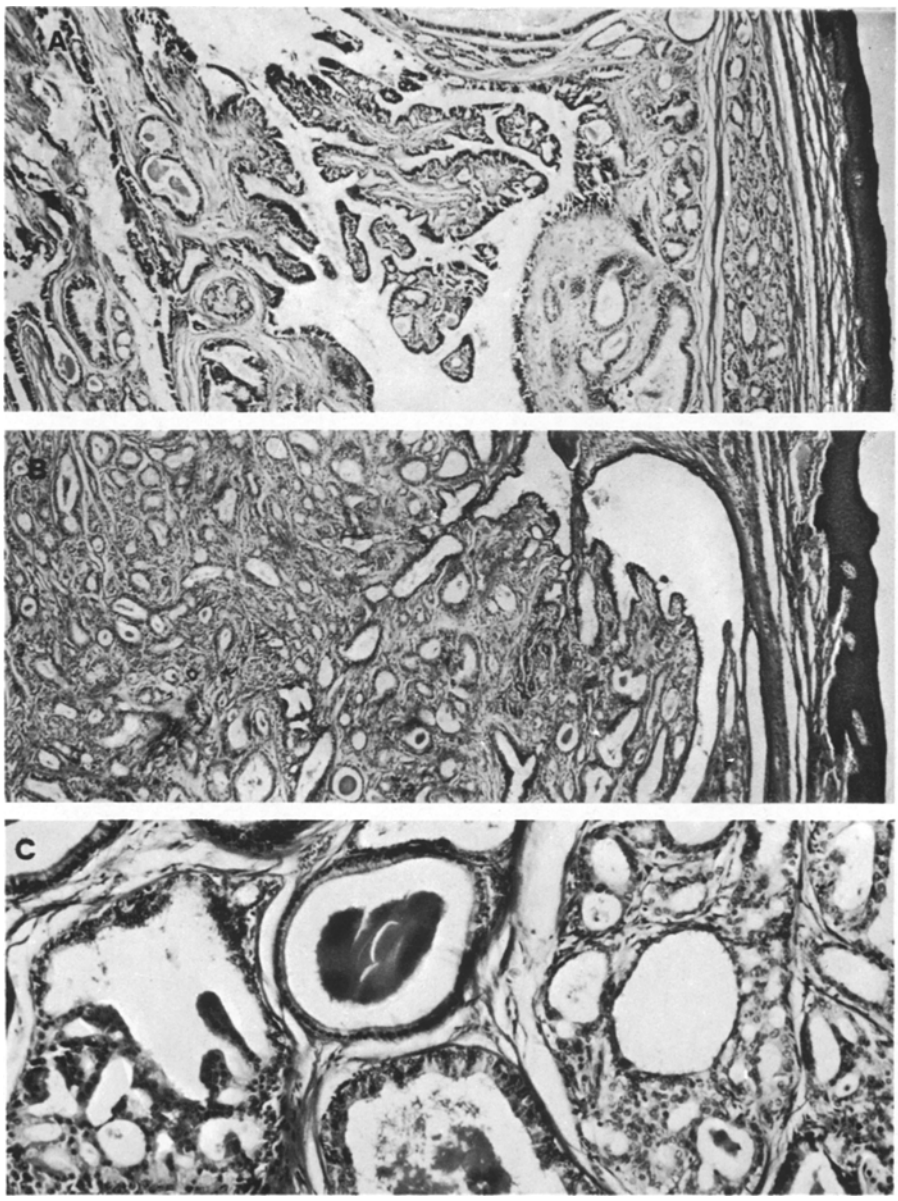

Fig. 2a–c. Adenoma of apocrine anal glands with papillary (**a**), tubular (**b**), and cribriform (**c**) patterns. All H & E. Magn. **a** and **b** ca. ×40, **c** ca. ×100

Histologically (5332/78), the specimen is covered partially with rectal mucosa and partially with anal squamous epithelium. In a fibrous stroma there is a small cyst (Fig. 1a) containing homogeneous PAS-positive material. The epithelial lining is double-layered, the inner layer showing active secretion with some "hob-nail" cells (Fig. 1d) characteristic of an apocrine mode of secretion. The diagnosis of an apocrine hidrocystoma was made.

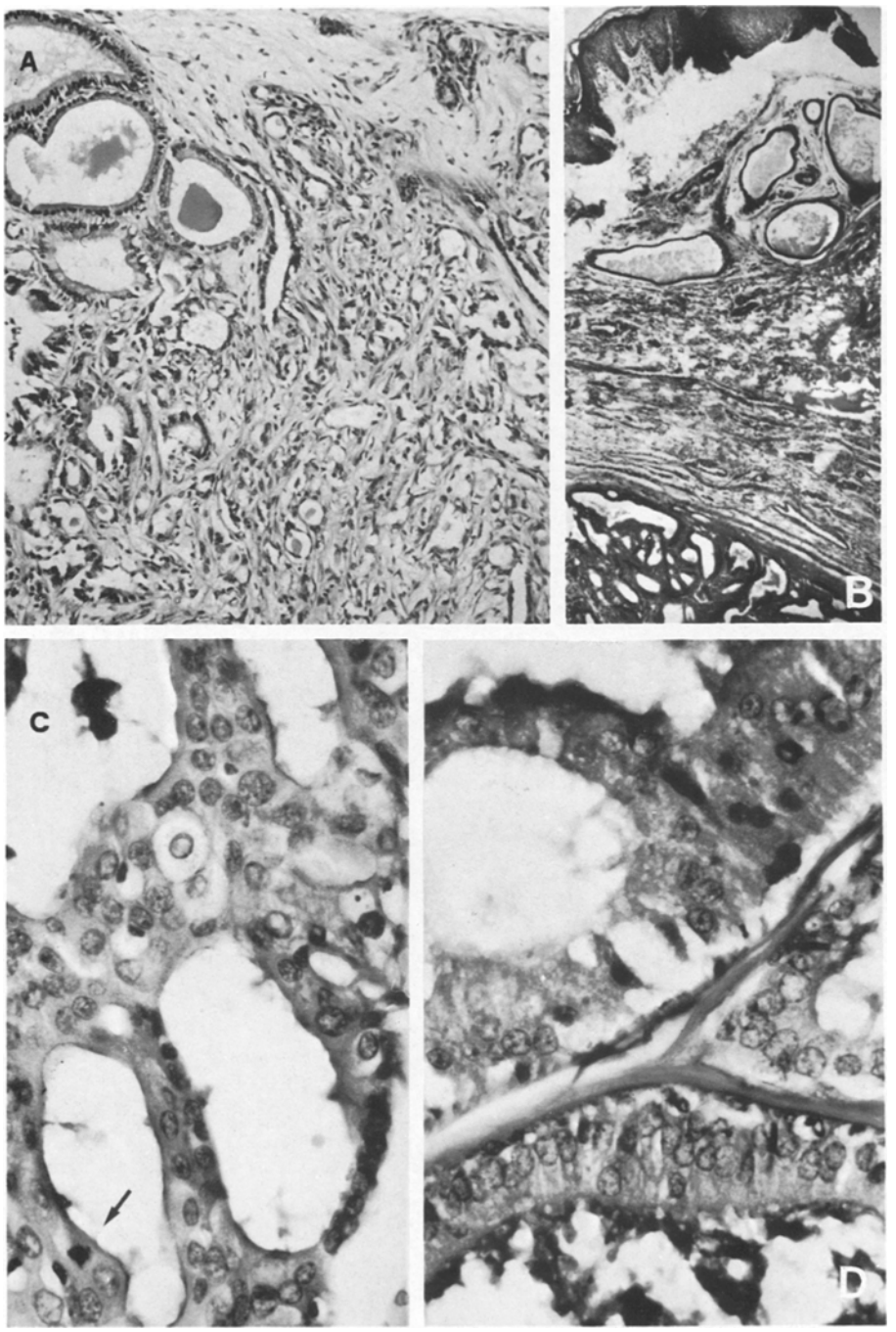

Fig. 3a–d. Adenoma of apocrine anal glands. Low differentiated tubular areas may occur (**a**). Near the margin of excision a small complex of cystic anal glands (**b**). Only in well differentiated areas can the apocrine mode of secretion be recognized (**d**). Less differentiated structures like the cribriform area in **c** may show anisonucleosis and single mitoses (→). **a–c** H & E, **d** PAS-Hematox. Magn. **a** ca. ×100; **b** ca. ×40; **c** and **d** ca. ×400

2. A 32-year-old woman with abdominal discomfort was first seen by a gynecologist who felt a mass on digital rectal examination. At operation a cylindrical submucosal cyst was discovered about 6 cm above the external anal ring. This could only incompletely be removed and during the procedure was damaged, releasing some opaque grayish fluid. Similar material has been described by Mehregan (1964) in comparable dermal lesions. Histologic examination (6074/78) shows a septate cyst, surrounded by connective tissue and bundles of anal muscle. Rudimentary papillary formations and scanty homogenous PAS-positive content are found (Fig. 1b). The epithelial lining is partially transitional, partially double-layered cylindrical with apocrine decapitation secretion (Fig. 1c). Single vesicular mucoid glandular tubules are present in the neighbourhood of the cyst, similar to those described by Hamperl (1974).

3. As incidental finding in three different hemorrhoidectomy specimens cystic structures of comparable size and epithelial content to those described above were discovered. The specimens were from women aged 39, 45, and 65 respectively.

4. During a gynecologic tumor-screening examination a pendulant mass with a short stalk was found in the anal ring of a 46-year-old woman, who reported difficulty during defecation. After surgical removal the specimen (2641/78) measured 2.5 cm in diameter, had an epidermal surface and showed some mucinous material on the cut surface. Histologically, the lesion is a well encapsulated adenomatous neoplasia of great structural variation forming cystic, papillary and tubular patterns, all embedded in a fibrous stroma (Figs. 2 and 3). Occasionally a cribriform pattern is evident (Fig. 2c). The tubular and cystic areas contain a varying amount of PAS-positive mucinous secretion. In some of the tubular and most of the cystic structures an apocrine mode of secretion can be demonstrated (Fig. 3d). Less differentiated areas are often lined by a less specialized epithelium, sometimes with only abortive mucous secretion (Fig. 3c). There is moderate variation in nuclear size and shape and rare mitotic figures can be found (Fig. 3c). Within the stalk near the margin of excision a small glandular, obviously normal complex with cystic dilatation is seen (Fig. 3b). Comparable observations are reported in the literature (cf. Meeker et al., 1962) in the vicinity of apocrine adenomas from other sites, apparingly indicating the hamartomatous nature of the lesions. The diagnosis of a predominantly tubular adenoma of apocrine anal glands was made.

Discussion

A previous report on anal gland cysts by Hamperl (1974) discussed morphogenic aspects and the scanty literature. However, there was no analysis of their mode of secretion or of their relationship to similar cystic formations occurring in the skin, particularly on the head, which are called hidrocystomas since the first description by Robinson (1893) and which are, similarly to the cystic formations described here, lined with an apocrine epithelium, if they are derived from apocrine glands. It is a matter of dispute, whether these clinically tumor-like lesions are retention cysts, as assumed by Hamperl (1974), or whether they represent neoplastic conditions. Mehregan (1964) argues, that if there is an active secretion of the epithelium they are better classified as a cystadenoma. Smith and Chernosky (1974) accept this view at least in those lesions forming papillary projections, which are not a constant finding and only rudimentary in hidrocystomas.

The neoplastic character of the second type of anal gland lesion described here is beyond dispute. Cooper and McDonald (1944) were the first to describe an anal tumor fitting the description of a hidradenoma given by Pick (1904). The similarity with adenomas of sudoriferous glands occurring in the genital

area, especially the vulva, is obvious, and the apocrine epithelial lining demonstrates the basic relationship.

Traditionally, the adenoma of apocrines weat glands is described as hidradenoma papilliferum. In the largest collection of cases (Meeker et al., 1962) 68% of tumors of this type showed a predominantly papillary pattern, but in every case tubular formations were also present. Tubular and papillary pictures of equal extent were noticed in 22% of the cases, and in 11% a predominantly tubular pattern was found. Thus Teloh's (1954) claim that hidradenoma papilliferum is merely the most common variant of apocrine gland adenoma seems justified.

In the series of Meeker et al. (1962) 22% of the cases (i.e., 14) originated from the anal glands. Apart from the 4 cases described by Teloh (1954) the rest of the literature consists only of single case reports (Cooper and McDonald, 1944; Cox, 1954; Rosenberg and Skerret, 1955; Shenoy, 1961). All reported adenomas were seen in women. Although our series of anal hidrocystomas is too small for a similar definitive statement, it is of interest that they too all occurred in women. The cause for this sex predilection is unknown and the fact that apocrine odoriferous glands are more numerous in females cannot be the only explanation. As this type of gland matures first after the onset of puberty, tumors would be unlikely to develop before some time after this, and no case is recorded where a tumor has been discovered before the age of 30.

In some cases of apocrine adenoma (Teloh, 1954; Meeker et al., 1962), particularly in tubular areas, foci of varying epithelial differentiation and occasional mitoses were described, similar to that seen in our case. These appearances were mistakenly interpreted as indicating malignancy in the earlier literature (cf. Meeker et al., 1962). The pattern developed in some areas of the adenoma described here would suggest a highly or even moderately differentiated adenocarcinoma, if one did not see the entire lesion and its intact borders. Quite similar pictures are formed in apocrine sweat gland carcinomas of other sites on the body, especially in the axilla (see Warkel and Helwig, 1978).

It should be emphasized therefore, that in carcinomas of the apocrine anal glands none of the various patterns of differentiation seen here have been described. Such carcinomas grow either in squamous or basaloid patterns or have frankly invasive, poorly differentiated collodial or mucoepidermoid appearances (Bigelow, 1976). The only case in the more recent literature interpreted as malignant change in a hidradenoma papilliferum is that reported by Shenoy (1961), which, on consideration of the case history and the photomicrographs is more likely to be an anal squamous cell carcinoma developing adjacent to a papillary adenoma of anal glands.

References

Bigelow, B.: Cancer of the anal and perianal areas. In: Cancer of the skin, Andrade, R., et al. (eds.). pp. 1394–1408. Philadelphia-London-Toronto: W.B. Saunders Comp. 1976

Cooper, W.L., McDonald, J.R.: Adenoma of apocrine sweat glands (hidradenoma) of the anal canal. Arch. Pathol. **38**, 155–157 (1944)

Cox, E.A.: Hidroadenoma of the perianum. Am. J. Surg. **88**, 789–792 (1954)
Hamperl, H.: Über "Analcysten". Virchows Arch. A Path. Anat. and Histol. **363**, 175–178 (1974)
Meeker, J.H., Neubecker, R.D., Helwig, E.B.: Hidradenoma papilliferum. Am. J. Clin. Path. **37**, 182–195 (1962)
Mehregan, A.H.: Apocrine cystadenoma. Arch. Dermatol. **90**, 274–279 (1964)
Pick, L.: Über Hidradenoma und Adenoma hidradenoides. Virchows Arch. Path. Anat. **125**, 312–364 (1904)
Robinson, A.R.: Hidrocystoma. J. Cutan. Genitourin. Dis. **11**, 293–303 (1893)
Rosenberg, J., Skerrett, P.W.: Hidradenoma of the anus. Am. J. Surg. **89**, 1081–1083 (1955)
Shenoy, Y.M.V.: Malignant perianal papillary hidradenoma. Arch. Dermatol. **83**, 965–967 (1961)
Smith, J.D., Chernosky, M.E.: Apocrine hidrocystoma (Cystadenoma). Arch. Dermatol. **109**, 700–702 (1974)
Teloh, H.A.: Apocrine adenoma of the anus. Cancer **7**, 367–372 (1954)
Warkel, R.L., Helwig, E.B.: Apocrine gland adenoma and adenocarcinoma of the axilla. Arch. Dermatol. **114**, 198–203 (1978)

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