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Hidradenitis Suppurativa: Diagnosis and Surgical Management of Perianal Manifestations

By RAYMOND J. JACKMAN, M.D.[1]

Rochester, Minnesota

"HIDRADENITIS" is derived from the Greek "hidros", which means "sweat", and "adenos", meaning "gland". It is marked by the development of one or more cutaneous shot-like nodules which enlarge to the size of a pea and undergo softening and suppuration, with subsequent discharge. It is characterized by the formation of abscesses, sinuses and undermined ulcerations. The sites are the cutaneous surfaces of the body where the apocrine sweat glands are located, namely, the axillary, sub-mammary, inguinal, genital and perianal regions.

Confusion in terminology.—Until 1939, when Brunsting [2] published a paper in which he pointed out that the disease is more common than generally appreciated and has been obscured under such names as "furunculosis of the axillas and buttocks", "abscess", "fistulous disease of the buttocks", "pyoderma" and "nonspecific granuloma", the diagnosis of hidradenitis suppurativa was rarely made. In fact, the name had been applied to the disease almost three-quarters of a century before Brunsting's paper, by a French surgeon, Aristide Auguste Verneuil [6], who recognized by clinical observation alone the association of the abscesses with the location of the apocrine sweat glands.

[1]From the Mayo Clinic and Mayo Foundation, Rochester, Minn. The Mayo Foundation, Rochester, Minnesota, is a part of the Graduate School of the University of Minnesota.

Pathogenesis.—Although it is almost universally accepted that hidradenitis suppurativa is related in some way to the apocrine sweat glands, there are those [1] who feel that the involvement of the glands is coincidental and secondary. Other than the presence of the apocrine glands, the so-called "sites of predilection" might be explained by lack of hygiene and by accumulation of moisture and generation of heat and friction in areas in which skin surfaces normally appose. It is also generally accepted that the disease has some relationship to endocrine activity and that persons who have hidradenitis suppurativa also have an acne diathesis.

Lending support to the premise that the apocrine sweat glands are primarily involved is the observation that the anal canal itself, which does not have any hair follicles and consequently no apocrine glands, is rarely involved by hidradenitis suppurativa. If the anus is involved it is an extension from a perianal abscess.

Sites of involvement.—In 388 patients studied by McQuarrie and myself [3], the most common site was the axilla which was involved alone or in combination with other areas in 278 (72%) of the group. In these 278 patients the axilla alone was involved in 178 instances.

The second common site was the perianal region; this was involved in 125 patients (32%) of the group. In about two-thirds of this group (84 patients) there was coexisting single or bilateral axillary involvement and because of this the diagnosis of the perianal disease was rather simple. In the 41 patients in whom there was perianal involvement alone or in combination with scrotal or labial involvement, the clinical diagnosis was sometimes wrong; the correct diagnosis could be made only at operation. 4 of these 41 patients were proved at operation to have extensive anal fistulas with a primary source in the anus and multiple secondary openings. Several other patients had undergone an unsuccessful operation for pilonidal cyst disease or an anal fistula which actually proved to be hidradenitis suppurativa. Therefore, at least some so-called recurrent pilonidal cysts or anal fistulas are recurrent or new areas of hidradenitis suppurativa. Similarly, some of the "difficult-to-cure anal fistulas" in which an anorectal source is not found are instances of hidradenitis suppurativa.

The mammary area was involved alone or in combination with other sites in 32 patients (8%). The groin was involved alone or with other parts in 92 patients and the back of the neck in 44 patients.

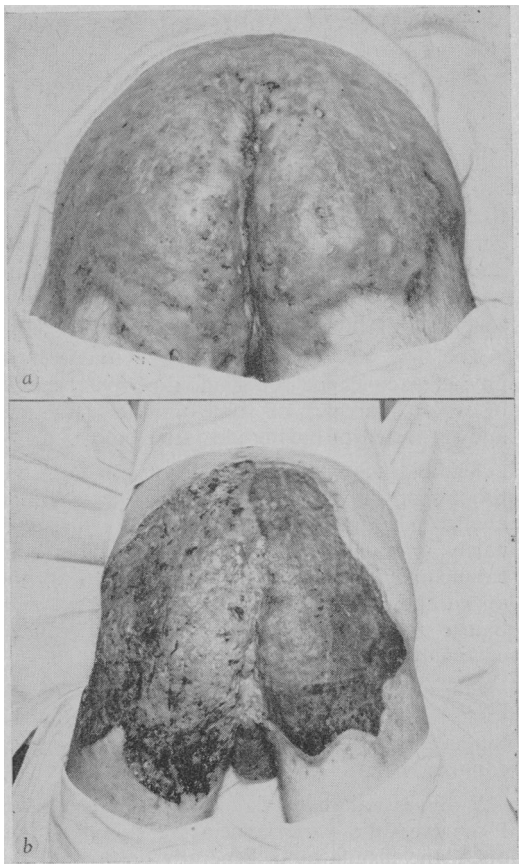

FIG. 1.—A, Extensive hidradenitis suppurativa, with numerous sinuses, undermining and induration. B, The same patient after excision of involved area and before skin grafting.

Age at onset.—The commonest age of the patient at the onset of the disease is within the second decade, meaning the age of puberty. This suggests that further investigations in the field of endocrinology are indicated.

Management.—In general, once the disease is established, non-surgical treatment has been ineffective. Various vaccines, injections of foreign proteins, antibiotic agents, roentgen-ray therapy, ultraviolet or infra-red light and the like have been tried but for the most part have been unsatisfactory.

Early recognition and early operation are important to prevent the disease from spreading. When lesions are small, incision and drainage usually are considered to be adequate, but in our opinion it is better, even when the lesions are in an early stage, to excise them widely and leave the wound open. These measures help to prevent recurrences. When lesions are long-

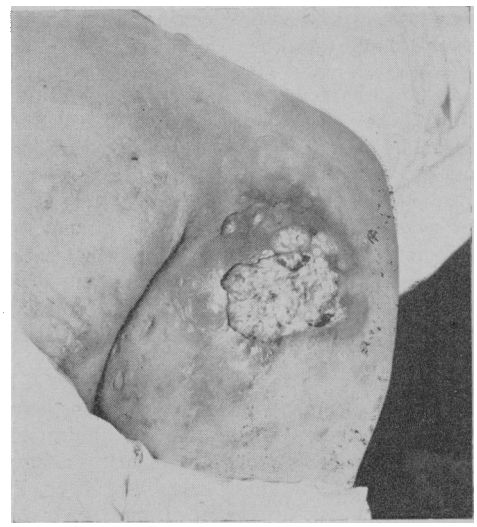

FIG. 2.—Long-standing hidradenitis suppurativa with superimposed fungating, low-grade squamous cell epithelioma. This was treated by wide local excision and subsequent skin grafting.

standing and extensive, with undermining and deep subcutaneous involvement, wide excision is carried down to healthy tissue (Fig. 1A), leaving an open wound. This constitutes the treatment of choice. After a week or possibly two weeks, when all infection has subsided and granulation tissue has started to form, split-thickness skin grafting is carried out (Fig. 1B). The area should be immobilized by some type of mould, to ensure a successful graft take. During this immobilization a minimal-residue diet is employed to avoid bowel movements for several days. Despite the perianal zone being a potentially infected area, various types of grafting are feasible and highly successful.

Squamous cell epithelioma in hidradenitis.—Several reports of the development of cancer in or near chronic draining sinuses are in the literature [4]. Generally, the sinuses will have been present for many years before the carcinoma appears. In the 125 cases of perianal or gluteal hidradenitis suppurativa previously mentioned, 4 squamous cell epitheliomas were found superimposed on the hidradenitis (Fig. 2). All 4 patients had had the disease for many years (nineteen to thirty-two years). The epitheliomas all tended to be large, and when they were first noted by the patient they were considered to be part of the inflammatory process, so that a year to four years elapsed before medical help was sought. 3 of the 4 lesions were grade 1 and the fourth was a grade 2 squamous cell epithelioma. This last patient eventually died from wide-

spread metastasis. Wide local excision appeared to have cured the other 3 patients.

Incidence of recurrence.—Satisfactory follow-up information was obtained from 38 of the 64 surgically treated patients studied by Anderson and Dockerty [1]. 8 (21%) of the 38 had no subsequent symptoms. 12 had mild symptoms but needed no further surgical treatment. The remaining 18, or almost half, of the patients did have further difficulty necessitating further surgical intervention; most of them presented new areas adjacent to the previously treated sites.

Measures to help prevent recurrences.—The following measures will, in our opinion, help to prevent further difficulty with hidradenitis suppurativa: (1) The most important is adequate initial excision, meaning wide and deep local excision by block dissection of areas of involvement. (2) After wide excision an anti-inflammatory dose of roentgen rays of 200 to 300 roentgens should be given in case some areas of inflammation remain [5]. The practice of administering roentgen-ray therapy with the objective of "drying up" or inactivating the apocrine glands has been largely abandoned because the dose required is so high that it may be dangerous. (3) Scrupulous daily hygiene in the sites of predilection, is carried out with detergent soaps and antiseptics.

Conclusions.—(1) Although the incidence of further difficulty with hidradenitis suppurativa, meaning new areas of involvement or areas of recurrence, is relatively high, surgical excision of involved sites is the best treatment for patients with this disease. (2) When hidradenitis suppurativa involves only the perianal, gluteal or sacral area, a problem in differential diagnosis between anal fistulas and pilonidal disease arises. Sometimes this problem can be settled only by examination with the patient under an anesthetic. (3) Although the incidence of complicating squamous cell epithelioma in hidradenitis suppurativa is not high, the possibility of the lesion must be considered, particularly when hidradenitis suppurativa is long-standing.

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Non-specific Anal and Perianal Ulceration

C. PATRICK SAMES, M.S., F.R.C.S.[1]
Bath

I AM prompted to give this contribution because non-specific perianal ulceration appears to receive scant recognition in the literature. It defies treatment so it has become rather the ugly duckling of proctology. These remarks are based upon experience of 3 problem cases under my care in the last few years.

The first was a woman referred to me by a physician colleague for sigmoidoscopy, because of severe diarrhœa of some months' standing, and suspected of having ulcerative colitis.

Sigmoidoscopy was normal to 30 cm., but proctoscopy revealed two shallow ulcers in the anal canal, together with a skin tag which was partly granulation tissue; and a shallow subcutaneous fistula-in-ano. The whole had the appearance of tubercle, with the rather weak bluish unhealthy granulations, the undermined edges and the thin seropustular discharge.

She also had peri-articular thickening of the joints of the hands and wrists, ulcers on the tongue and palate, and a phlyctenular type of conjunctivitis.

With a tentative diagnosis of tuberculosis I removed the partly ulcerating skin tag, laid open the fistula and generally improved the drainage.

The biopsy report was non-specific granulation tissue with no evidence of tuberculosis. The E.S.R. was persistently raised to over 80 mm. in one hour (Westergren) and the hæmoglobin fluctuated around 45–55%. Barium enema and barium meal were negative, without evidence of ulcerative colitis or Crohn's disease. Several searches were made for L.E. cells; the Rose's test was always negative and serum Wassermann and Kahn reactions likewise.

The ulceration of the anal canal and perianal skin refused to heal despite every effort. Exposure to ultraviolet and infra-red light made no impression. Several searches were made for unusual anaerobic organisms, for amœbæ, and *Giardia lamblia*, all without success. Despite cortisone therapy, heavy doses of vitamins and a course of mepacrine, the condition worsened.

In an attempt to arrest its slow but relentless advance I resorted to diathermy excision, on the lines of treatment recommended for symbiotic synergistic gangrene. This was followed by an appreciable reactionary hæmorrhage, which she

[1]Surgeon to Royal United Hospital, Bath.