Case Report

A Life-Threatening Multilocalized Hidradenitis Suppurativa Case

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Abstract and Introduction

Abstract

The patient was a 38-year-old man. He had been suffering from hidradenitis suppurativa (HS) for approximately 20 years. He had active lesions at both axillae, hip, scrotum, and perineum, and inactive lesions located behind the ears, lower abdomen, and posterior neck. He was monitored and treated at different branches; he continuously used antibiotics and was given steroids at times. Antibiotic resistance developed subsequently. His general situation was bad; vital signs were poor; and he was in a state of sepsis and preshock, so this case was regarded as life-threatening.

Total excision was performed first on the lesion at the right axilla, then on the lesion at the left axilla, and the parascapular fasciocutaneous flap was reversed. A skin graft was applied to the triangular defect on the scapula. No relapse occurred. Then the lesions at the hip were managed. Broad excision was used twice with the patient under general anesthesia; because the lesions spread to the retrococcygeal and gluteal muscles, coccyx resection and partial gluteal muscle resection were implemented. The defect was eliminated with a progressive flap. At the intergluteal sulcus, small lesions emerging at the median line were debrided with the patient under local anesthesia, and together with secondary recovery, the disease was completely managed. Lesions at the perineum and scrotum and at both inguinal areas were broadly excised and grafted. No lesion has relapsed so far. One year later, Hodgkin's lymphoma was diagnosed, and the patient was treated with chemoradiotherapy easily, because there was no infective focus. The disease is in remission now. The patient weighs 110 kg, is healthy, and is working again.

Introduction

HS is a disease that is characterized by intensive development of sclerosis due to abscess, sinus and fistula formation, and fibrosis as a result of a chronic infection of the apocrine sweat glands. The disease can be seen at the scrotum, perineum, perianal region, hip, axilla, periumbilical region, and breast areola -- any area that has apocrine sweat glands. It has a typical appearance (Figure 1). The disease was first defined by Valpeau in 1839. The incidence and prevalence of the disease are not known; however, in some studies, a 1% incidence is declared.[1-3] This disease occurs more frequently in women.[4] Hereditary factors, obesity, and sports are regarded as predisposing factors. The disease is not seen before puberty, because the apocrine sweat glands become active with the effect of the sex hormones, which are inactive during prepuberty.[2,5] However, plantar HS or palmoplantar HS can be seen in children.[6,7] The facilitation of bacterial translocation due to the obstruction of sweat glands and abnormal excretion of sweat glands as a result of insufficient or inaccurate development is regarded as the cause of HS. Recent research indicates that these patients have neutrophil dysfunction as well.[8]
In some patients, the disease becomes complicated, and serious antibiotic resistance develops. Because of antibiotic resistance, infections cannot be controlled, and large abscesses may develop. Septic complications may develop that necessitate hospitalization of the patient and administration of broad-spectrum antibiotics. This period can last 10 to 15 years. In addition, in patients with HS lymphedema, complications such as contracture formation at the region of the lesion, squamous cell cancer development, restriction of movement, urethral or rectal fistula development, anemia, arthritis, amyloidosis, renal failure, and interstitial keratitis may develop. On the other hand, in some patients the disease may regress spontaneously.

Fatal complications of HS are rare; however, especially in multilocalized cases, serious morbidity occurs, affecting a patient's social and professional life.

The Patient

In December 1999, a 38-year-old male patient resorted to SSK (Social Security Institution) Ankara Training Hospital General Surgery Polyclinic 2 with complaints of pain at the hip, flow of pus, and weight loss. The patient was using a pad; at physical examination when the pad was opened, there was a putrid smell, and all around the hip, there were multiple, flowing fistulas and sinuses; intensive sclerosis; and a swelling causing fluctuation in an area that was close to the midline. There were also lesions at both axillas, behind the ear, the periumbilical region, the scrotum, the perineum, and the inguinal region. Except for the lesions located behind the ears and the periumbilical region, all the lesions were active (Figures 2 and 3). His vital signs were poor: blood pressure was 80/60 mm Hg, pulse rate was 124 beats per minute, body temperature 38.4°C. CBC count results showed 21,100; hemoglobin, 9.6 g/dL. The patient had lost 35 kg, with actual body weight of 81 kg (normal weight, 116 kg.) He was in a state of sepsis and preshock. Shortly, his case was considered to be life-threatening.
Superficial ultrasonography showed a gluteal abscess. The abscess was drained immediately; approximately 3 L of abscess content was discharged. Samples were taken from abscess content for examination. Blood culture results were negative; however, in the abscess content, culture coagulase-positive *Staphylococcus aureus* was found, with resistance against antibiotics, including tetracycline, ciprofloxacin, ornidazole, gentamicin, neumamycin, trimethoprim-sulfamethoxazole, and clarithromycin. Abscess content was washed with copious isotonic NaCl solution and rifampin plus hydrogen peroxide mixture was instilled. The same type of treatment dressing was applied to the patient every day, and without using any antibiotics, the infection was under control in 1 week.

HS had been diagnosed 20 years previously; the lesions became more active, more widespread, and more painful with increased flow in the recent 5 years. Because of the putrid smell associated with infection in addition to his symptoms, the patient was unable to sustain his professional and social life. The patient, monitored by...
dermatologists, was continuously given antibiotics; he also used isotretinoin for 1 year and 60 mg/day of steroids for 3 months. In another hospital, excision and primary suture had been applied to the lesions at axilla; however, the disease relapsed on the twelfth postoperative day.

The patient was briefed about all major and minor risks, including relapse, and surgery was performed on the right axilla 18 April 2000. The disease was accepted just as a malignant lesion, and surgical edges were formed by making sound edges of at least 2 cm out from the lesions; we reached the fascia vertically, and a very wide excision was made. Then we entered the axillary fossa; the axillary vein was disposed; and some reactive huge (3 x 4 cm) lymph nodes were extracted. Then via plastic surgery, a parascapular fasciocutaneous flap was reversed and the defect was covered. The triangular defect on scapula was covered by a skin graft. The operation was concluded with the placement of a drain (Figures 1, 4, and 5).

**Figure 4.**
Axillary fossa and axillary vein disposed.

**Figure 5.**
Skin graft covering the defect on the scapula.
During the postoperative follow-up, there were no problems, no infection, and no flap necrosis. The skin graft showed compliance and no relapse has occurred within 2 months. Therefore the same surgery was performed on the left axilla after 3 months, and the same result was achieved. Six months after this operation, the lesion at the hip, which was the most difficult, most complicated, and most widespread, was managed with surgery (Figure 2). Again the lesions were lined to have sound edges; however, unlike the lesions in the axillas, these were going beyond the fascia and invaded the gluteal muscles, and the lesion at the intergluteal sulcus reached up to the retrococcygeal area. Approximately 70% of the hip was excised together with fascia and some of the gluteal muscles (Figures 6 and 7). There was only 2 cm of distance to the anal channel; upper parts of the external sphincter were also affected by the disease and partially resected. The wide defect was covered with a rotational fasciocutaneous flap. However, the defect at the midline was grafted by means of a skin graft (Figures 8 and 9). During the postoperative follow-up examinations, areas except for the midline have been completely covered; however, the graft at the midline was rejected and flow occurred. Regrafting was tried, but failed again. Considering that it might have been caused by a retrococcygeal sinus tract, coccyx resection, curettage, and skin graft were implemented 1 month later. In the follow-up examinations, the flow decreased but the graft was rejected again, and the scar was left to secondary healing. Over 6 months, partial excision and curettage were performed a few times with the patient under local anesthesia. As a result of these treatments, the lesion was completely eliminated (Figure 10). After the treatment, all the findings of chronic infection were gone, and that patient did not have anemia.

**Figure 6.**
Appearance of the scrotal region during the follow-up period.

**Figure 7.**
Left axilla.
Figure 8.
Right axilla.

Figure 9.
Wide excision applied, including fascia and gluteal muscles.
Figure 10.
Partial excision and curettage performed in the follow-up.

The last surgical procedure was performed on the area including the bilateral inguinal area, scrotum, and perineum, which was the most complicated area following the hip (Figure 3). Partial scrotum, perineum, and inguinal area excision plus primary repair were performed. On the fifth postoperative day, the sutures on the lateral edges of scrotum were detached, and a skin graft was applied on these areas. This area recovered completely in 2 weeks (Figure 11).

Figure 11.
Fifth postoperative day: Skin graft was applied after sutures on the lateral edges of scrotum were detached.
Discussion

The incidence, prevalence, and etiology of HS are not known. However, it is believed not to be rare.\textsuperscript{[4,10]} In an important portion of cases, there are small lesions that can be tolerated by the patient because they are not frequently active. Many patients can be misdiagnosed as having pilonidal sinus, folliculitis, or furuncle. For this reason, the disease can be widespread for many years and may lead to serious physical and social morbidity. The pain, restriction of movement, anemia, abscess flow, and smell caused by the infection may put an end to the social and professional life of the patient.

After the lesions at the axilla are deeply excised with both broad and partial lymphatic dissection, the wide graft that emerges should absolutely be covered by a flap to avoid restriction of movement. Different methods are defined.\textsuperscript{[10-12]} The method we used is a parascapular fasciocutaneous flap. On the other hand, a small triangular defect on the scapula is covered with a skin graft. The results are perfect. No restriction of movement is encountered (Figures 12 and 13).

\begin{figure}[h]
\centering
\includegraphics[width=\textwidth]{gluteal_region.png}
\caption{The appearance of the gluteal region at the end of the operative procedure.}
\end{figure}

\begin{figure}[h]
\centering
\includegraphics[width=\textwidth]{scapula.png}
\caption{The appearance of the scapula at the end of the operative procedure.}
\end{figure}
In the follow-up period there is no restriction of movement.

The characteristics of the lesions at the hip change according to the chronic pressure; while they should be limited to the fascia, they also affect the gluteal muscles, and they even reach up to the retrococcygeal area and cause the formation of rectal fistulas. Therefore, we think the lesions in this area should be excised widely because the lesions in this location are much larger prior to such complications. More importantly, the lesions at this area should be diagnosed earlier, we think early surgical intervention should be performed. Such an attitude will facilitate the surgical treatment as well. In the magnetic resonance images of the hip lesion of our patient taken 6 years previously, the diameter of the lesion was about one fifth of the size of the lesion when we operated. Treatment will get even more difficult as a result of the growth of the fistulas.

The lesions at the scrotum and perineum should be treated surgically because the lesions in this area are more painful, cause movement restriction, and carry the risk of urethral fistula. After total excision, the defect must be covered by a skin graft. The HS lesions in other sites may sometimes be treated medically.

The coexistence of HS with diseases, such as Crohn's disease, irritable bowel syndrome, Down syndrome, Grave-Hashimoto thyroiditis, arthritis, Sjögren's syndrome, and herpes simplex, is more frequent than that in the normal population. We detected hypospadias in our patient and at the end of the treatment, Hodgkin's lymphoma was diagnosed. We could not find any coexistence of hypospadias and HS in the literature. And, to our knowledge, there is no information in literature about the relationship between HS and Hodgkin's lymphoma.

**Conclusion**

HS is a curable disease. Besides the frequently recurring cases, newly emerging gluteal, scrotal, perineal, and perianal lesions should be operated on immediately because of higher rates of complications. Small lesions at the axilla, retroauricular, periumbilical areas, can be monitored medically. However, in cases of a rapid increase in development of lesions, we think early surgical intervention is a necessity.

Together with broad excision of the lesions and the use of a fasciocutaneous rotational flap, which is not tense, and skin grafts, the relapse ratios are quite low. All of the relapses are seen in the first 4 weeks, and with reoperation and broader excision, the problem can easily be solved. The fact that Hodgkin's lymphoma developed in our patient at the end of treatment meant that he could undergo complete, successful chemoradiation therapy. This was a great chance for this patient; however, that chance may not be present for all patients.

**References**

Abstract


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Readers are encouraged to respond to George Lundberg, MD, Editor of MedGenMed, for the editor's eye only or for possible publication via email: glundberg@medscape.net.

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