

건강한 성인에서의 오진하기 쉬운 포도구균성 열상 피부증후군의 치험례

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Staphylococcal Scalded Skin Syndrome in a Healthy Adult: Easy to Misdiagnose

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A 60-year-old male presented with a three-month history of redness and swelling on his left little finger. His medical history was not informative. Wound culture revealed methicillin-resistant *Staphylococcus aureus*. After vancomycin administration, the skin lesions became worse and whole body bullae and desquamation occurred. This was initially suspected to be a drug eruption; thus, we switched antibiotics from vancomycin to teicoplanin. However, biopsy revealed Staphylococcal scalded skin syndrome (SSSS). After several days, generalized skin symptoms improved. The patient recovered and is in good physical health without recurrence six months later. We describe a localized form of SSSS, which is very rare in healthy adults. Consequently, there is a high risk of misdiagnosis. Thus, we report a rare case of SSSS in a healthy adult and the importance of early histological examination for accurate diagnosis.

Key Words: Cellulitis, Drug eruption, Staphylococcal scalded skin syndrome, Vancomycin

Staphylococcal scalded skin syndrome (SSSS) is a generalized exanthematous disease with symptoms including cutaneous tenderness and broad superficial blistering and denudation. SSSS is caused by bacterial exotoxins A and B, which are released from *Staphylococcus aureus* and cause detachment of desmoglein within the epidermal layer¹. Epidermolysis usually takes place between the stratum spinosum and granulosum¹.

Although it is more commonly seen in infants and chil-

dren, SSSS can also occur in adults¹, in whom its rare appearance is usually associated with kidney failure, malignancy or immunosuppression. The case described herein is of interest because of its onset in a healthy adult and its mild clinical course.

CASE REPORT

A 60-year-old male presented with a three-month histo-

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ry of redness and swelling on his left little finger. He had a history of local trauma with sharp metal three months prior. Since then, the redness and swelling had slowly extended to the whole finger. He had undergone antibiotic treatment at a local clinic without symptom improvement. His medical history was not informative. In order to differentiate from bacterial tenosynovitis, physical examination was performed. On the initial physical examination, hand range of motion was in normal range. His finger did not hold in flexion position and there was no pain aggravation when passive extension. At the time of hospitalization, his body temperature was 37.4°C. Erythema with



Fig. 1. Redness and mild swelling on the left little finger.



Fig. 2. There were no radiologic abnormalities.

generalized small erythematous punctiform eruptions were present on the little finger (Fig. 1) and there were no radiologic abnormalities (Fig. 2). Routine laboratory tests did not reveal any abnormalities, other than slight inflammatory signs: C-reactive protein, 0.78 mg/dL and erythrocyte sedimentation rate, 40 mm/hr. The patient was negative for HIV antibody and other immunological examinations did not reveal any abnormalities. He was hospitalized and wound culture was performed. Empirical intravenous antibiotics (cephazedone+nesomicin) were used for cellulitis treatment. On hospital day 4, wound culture results indicated methicillin-resistant *Staphylococcus aureus* (MRSA). Skin symptoms had not improved (Fig. 3). Therefore, the antibiotic regimen was switched to vancomycin. After 3 days of vancomycin use, the patient developed a generalized fever, skin bullae on the trunk (Fig. 4), and desquamation of the top layers of skin, clear yellowish discharge on the left hand. Initially, we assumed this indicated a drug allergy and treated the patient with intravenous anti-histamines and steroids; however, there was no response. Five days later, skin desquamation and discharge had worsened (Fig. 5), with multiple bullae and desquamation extending to the whole hand. Therefore, to rule out drug eruption, such as Stevens-Johnson syndrome (SJS) related to vancomycin, as a possible cause, we performed skin biopsy. Mean-



Fig. 3. Hospital day 4. Despite of 4 days antibiotics treatment, skin symptoms did not improve.



Fig. 4. Hospital day 7. After 3 days of vancomycin use, multiple bullae on the whole body appeared. (A) Left lateral chest. (B) Back and sacral area.



Fig. 5. Hospital day 12. Skin desquamation and discharge on left hand were even worse. (A) Dorsal side. (B) Palmar aspect.

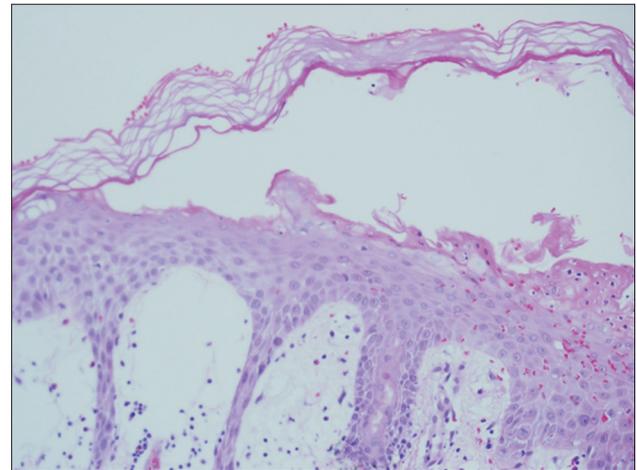


Fig. 6. Skin biopsy result. Subcorneal splitting of the epidermis with superficial epidermal necrosis is seen. There are few inflammatory cells within the bulla cavity and there is only slight damage to the underlying epidermal cells. It suggests staphylococcal scalded skin syndrome (H&E, $\times 100$).

while, antibiotic treatment was switch to teicoplanin. After several days, generalized skin symptoms improved progressively, and skin biopsy results indicated SSSS (Fig. 6). Since the patient was already treating with strong antibiotics to cover the MRSA, we observed the progress while continuing the same treatment. He was responding to antibiotic treatment and showed improved symptoms. Patient discharged from the hospital and returned for out-patient follow-up. He is in good physical health without recurrence 6 months later (Fig. 7).

DISCUSSION

SSSS is the clinical term that used for a spectrum of blistering skin diseases induced by exfoliative toxins type A or B from *S. aureus*. The severity of SSSS varies from a few blisters in localized SSSS to severe exfoliation affecting most of the body surface in generalized SSSS.

SSSS can occur at any age but it is the most common in children less than 5 years of age^{1,2}.

As opposed to pediatric patients who are usually in otherwise good health when they develop SSSS^{3,4}, virtually all adult cases of SSSS occur in those who are immunocompromised due to chronic renal disease, HIV infection, malignant neoplasms, chemotherapy, intravenous drug abuse or diabetes mellitus. Similar to pediatric patients, adults with SSSS demonstrate a fever, erythema, bullae formation and desquamation of the skin. Possibly as a result of underlying morbidities, adults with SSSS have a mortality rate of 40%-63%^{5,6}.

SSSS typically needs to be distinguished from drug eruption, as skin bullae and desquamation are common clinical symptoms. In particular, severe drug eruptions, known as toxic epidermal necrolysis (TEN) or SJS, need

to be distinguished from SSSS. In adults, both TEN and SSSS have a high mortality rate and require different treatment. Clinician awareness of SSSS is important in early diagnosis and treatment.

To differentiate SSSS from TEN, findings suggestive of SSSS include striking cutaneous tenderness, positive Nikolsky sign, and lack of mucosal involvement. In TEN, ulcerated mucosa is common, but diagnosis is not possible from clinical features alone (Table 1)⁶. A biopsy is necessary for accurate differential diagnosis.

In this case, we first suspected as TEN because the patient was a healthy adult without renal failure. Although

the period of infection was long, there was no systemic skin symptom. Systemic symptoms—such as Nicolsky's sign—appeared after using vancomycin. However, we could not stop the antibiotics because of the possibility of SSSS. So we replaced the antibiotics with another things and performed skin punch biopsy immediately. As result of biopsy, SSSS was confirmed. And Bacterial culture result also revealed as MRSA. Consequently, the patient was cured by proper antibiotics usage.

Once diagnosed as SSSS, hospitalization, rehydration and intravenous systemic antibiotic therapy become the principle of treatment. As the review of the literature, *S. aureus* was isolated from skin lesions in many adult patients⁷. In case of our patient, MRSA was found in wound culture. There were some cases caused by methicillin-resistant *S. aureus* have been reported^{8,9}. Comprehensively, early wound culture to find the causative bacteria and using the suitable antibiotics are best treatment of choice. Early strong antibiotics usage can lead to good results.

This patient exhibited systemic skin vesicles, multiple bullae, desquamation of the left small finger and a slight increase in inflammation. We suspected drug eruption because of wound aggravation after 3 days of vancomycin treatment. We stopped vancomycin, which was thought to be the causative drug, and switched treatment to teicoplanin. We also performed a biopsy for differential diagnosis. Because of the diagnosis of SSSS on histological examination, antibiotics were continued.



Fig. 7. After discharge from the hospital with a 3-month follow-up clinical photo in the well-healed state. (A) Dorsal side. (B) Palmar aspect.

Table 1. Distinction from TEN and SSSS

Clinical features		TEN	SSSS
Usually seen	Adults		Young children, infants A few adults with renal failure
Mortality	25%-35% (up to 60%)		4% in children and infants (up to 63% in adult SSSS)
Blister coloration	White		Tan or Brown
Mucosal involvement	Yes		No
Affected skin layer	Epidermal-dermal junction (keratinocytes)		Subcorneal (desmoglein 1)
Pathophysiological mechanism	Unknown (suspicious delayed hypersensitivity reaction)		Bacterial exotoxin
Histologic findings	Dermal-epidermal blister with focal dyskeratosis Full-thickness epidermal necrosis		Subcorneal blister, splitting
Treatment	Elimination of an offending drug (usually an antibiotics)		Appropriate antibiotics usage to treat the underlying bacteria

TEN: toxic epidermal necrolysis, SSSS: Staphylococcal scalded skin syndrome.

Optimal treatment from drug eruption, such as SJS and TEN, is to stop the causative drug. However, treatment of SSSS requires continuing antibiotic therapy. Drug eruption and SSSS have similar symptoms but differing treatment. If these diseases cannot be differentiated, high mortality can occur in adult patients. Adults, whitish blister color, full thickness epidermal necrosis suggest that TEN rather than SSSS but the differential diagnosis of scalded-skin lesions in healthy adults can be difficult; thus, reliable histologic examination is particularly important in SSSS and TEN. It is very important that mortality rate is up to 35% high in TEN and 63% in SSSS. Early histological examination for correct diagnosis is the most important and that can lead to better result.

We think it may be meaningful report to predict the rare case of SSSS which was clinically mild course on healthy adult.

In this case, it is meaningful because the possibility of infection by exfoliative toxin B cannot be ruled out. When a patient present with generalized symptoms such as skin bullae and desquamation, drug eruptions are typically suspected. However, clinicians should consider SSSS in cases of *S. aureus* infection and be aware of the need for prompt biopsy to ensure an accurate and timely diagnosis.

CONFLICTS OF INTEREST

The authors have nothing to disclose.

REFERENCES

1. Fitzpatrick TB, Goldsmith LA, Katz SI, Gilchrest BA, Paller AS. Fitzpatrick's dermatology in general medicine. 8th ed. New York: McGraw Hill Medical; 2012. 2148-60.
2. Miyashita K, Ogawa K, Iioka H, et al. Adult case of Staphylococcal scalded skin syndrome differentiated from toxic epidermal necrolysis with the aid of dermoscopy. *J Dermatol.* 2016;43:842-3.
3. Aydin D, Alsbjørn B. Severe case of Staphylococcal scalded skin syndrome in a 5-year-old child - case report. *Clin Case Rep.* 2016;4:416-9.
4. Mishra AK, Yadav P, Mishra A. A systemic review on Staphylococcal scalded skin syndrome (SSSS): a rare and critical disease of neonates. *Open Microbiol J.* 2016;10:150-9.
5. Cribier B, Piemont Y, Grosshans E. Staphylococcal scalded skin syndrome in adults. A clinical review illustrated with a new case. *J Am Acad Dermatol.* 1994;30:319-24.
6. Badon HR, King J, Brodell RT, Byrd A. Distinguishing features: Staphylococcal scalded skin syndrome vs toxic epidermal necrolysis. *SKIN J Cutan Med.* 2018;2:135-9.
7. Patel GK, Finlay AY. Staphylococcal scalded skin syndrome: diagnosis and management. *Am J Clin Dermatol.* 2003;4:165-75.
8. Ito Y, Funabashi Yoh M, Toda K, Shimazaki M, Nakamura T, Morita E. Staphylococcal scalded-skin syndrome in an adult due to methicillin-resistant Staphylococcal aureus. *J Infect Chemother.* 2002;8:256-61.
9. Yamaguchi T, Yokota Y, Terajima J, et al. Clonal association of Staphylococcus aureus causing bullous impetigo and the emergence of new methicillin-resistant clonal groups in Kansai district in Japan. *J Infect Dis* 2002;185:1511-1516.

건강한 성인에서의 오진하기 쉬운 포도구균성 열상 피부증후군의 치험례

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60세 남성이 3개월간 좌측 새끼손가락이 붓고 그곳의 발적 증상을 주소로 내원하였다. 과거력 상 특이 소견은 없었고, 균 배양 검사상 methicillin-resistant *Staphylococcus aureus*가 검출되었다. 반코마이신을 투여 후, 피부 병변이 악화되면서 전신 수포와 낙설이 발생하였다. 우리는 처음에 약물 발진 의진 하에 항생제를 반코마이신에서 테이코플라닌으로 변경하였다. 하지만 피부 조직검사 결과는 Staphylococcal scalded skin syndrome (SSSS)으로 나왔다. 수일 후, 환자는 전신 피부증상 호전되어 퇴원하였다. 우리는 건강한 성인에서 발생한 국소적 양상의 SSSS에 대해 보고하고자 한다. 이는 오진하기 쉬운 증례로서, 정확한 진단을 위하여서는 조기에 조직학적 검사가 중요할 것으로 생각된다.

색인단어: 연조직염, 약물발진, 포도구균성 열상 피부증후군, 반코마이신

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