CASE REPORT

Teenage boy with high fever and rash: what could we be missing?

Gracinda Nogueira Oliveira,¹ Sofia Costa,¹ Filipa Dias Costa,² Teresa Dionísio,² Leonor Carvalho²

¹Department of Paediatrics, Hospital Pediátrico de Coimbra, Coimbra, Portugal
²Paediatric Intensive Care Unit, Hospital Pediátrico de Coimbra, Coimbra, Portugal

Correspondence to

Dr Gracinda Nogueira Oliveira, gracinenoliveira@gmail.com

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SUMMARY

A 15-year-old boy was admitted to a local hospital with high fever, generalised rash and a mild sore throat. He was started on intravenous flucloxacillin and 12 hours later develops a sustained low diastolic blood pressure (DBP), unresponsive to fluid volume expansion and cardiovascular support with dopamine. Intravenous clindamycin was added and transportation to paediatric intensive care unit arranged. Dopamine dosing was increased and norepinephrine infusion was added subsequently with immediate stabilisation of DBP. A sacrococcygeal pilonidal abscess was identified, requiring prompt surgical drainage. The microbiological culture of abscess material was positive for an enterotoxin-producing *Staphylococcus aureus* and *Peptostreptococcus magnus*. He was free of symptoms after 4 days. This case report summarises a potential severe complication of the pilonidal disease.

examination revealed hyperaemia without exudate or tonsillar hypertrophy. Glossitis was noted (figure 3). There was no relevant personal or family medical history.

INVESTIGATIONS

He was admitted, monitored, underwent blood sampling and promptly started on intravenous flucloxacillin for suspected streptococcal TSS (STSS). Investigations revealed a negative rapid streptococcal antigen test, a white cell count of $15.3 \times 10^9/L$ (normal $5-15 \times 10^9/L$) with raised neutrophil count of $14.3 \times 10^9/L$, a slightly low platelet count of $161 \times 10^9/L$ and haemoglobin of $13.6 g/dL$. C reactive protein was raised ($13.2 mg/dL$). Creatine phosphokinase level was within the normal range for age ($245 UI/L$). He had normal liver and kidney function tests, as well as adequate electrolyte levels. A blood culture was requested.

BACKGROUND

Pilonidal disease is a common debilitating condition often neglected by teenagers. This patient was admitted with a history of fever, rash and mild sore throat. He also had a 7-day history of sacrococcygeal pain that he disregarded and only complained to his parents on a single occasion. Even though parents initially denied ongoing cutaneous infections, since no lesions were ever seen, a careful questioning elicited the memory of that complaint. This patient could be given a more timely directed therapy if pilonidal disease had been suspected earlier.

By writing this case report, the authors intend to provide a reminder that neglected pilonidal sinus disease can lead to unusual presentations within the clinical spectrum of toxic shock syndrome (TSS), with potential life-threatening complications.

CASE PRESENTATION

A 15-year-old man presented to the emergency department of a local hospital on the third day of fever and first day of a generalised rash and a sore throat. On examination, he was febrile ($40^\circ C$), haemodynamically stable and had a generalised erythematous non-pruritic maculopapular rash predominantly in the trunk and lower limbs with very mild desquamation seen on the face and left lower leg and two confluent macules on the lower legs (figures 1–3). Bilateral conjunctival hyperaemia was present, with no exudates. The oropharynx

DIFFERENTIAL DIAGNOSIS

- ▶ STSS: high fever, generalised maculopapular rash, sore throat and pharyngeal injection are very suggestive, becoming the most likely diagnosis at this point, despite the absence of tonsillar exudate.
- ▶ Staphylococcal TSS: high fever, generalised maculopapular rash, oropharyngeal and conjunctival hyperaemia are suggestive, but neither skin/soft tissue infection nor additional symptoms were found at this point.

TREATMENT

As an inpatient, he continued to spike high temperatures and after 12 hours and two doses of flucloxacillin, low diastolic blood pressure (DBP) was noticed. At this point, physical examination revealed a temperature of $37.4^\circ C$, blood pressure of $103/33 mmHg$ (mean $58 mmHg$), heart rate of 103 beats per minute, capillary refill time $<2 s$, 15-point score on Glasgow Coma Scale, regular cardiac and pulmonary auscultation, abdomen was non-tender without palpable masses and the generalised exuberant rash persisted with the same distribution. A venous blood gasometry was performed with normal results (lactate level of $1.7 mmol/L$). Rapid volume expansion with $20 mL/kg$ of isotonic sodium chloride was administered intravenously (three bolus) with no DBP response, leading to the initiation of dopamine at $5 \mu g/kg/min$. A nasal swab for *Staphylococcus aureus* (SA) screening was

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Figure 1 Erythematous maculopapular rash seen on both legs, with confluent macules.

collected, and intravenous clindamycin was added to flucloxacillin, considering the empirical treatment of TSS. Low DBP persisted and transportation to a tertiary hospital was arranged. During transportation, he felt nauseous and vomited twice. He was admitted to paediatric intensive care unit (PICU) and kept on cardiorespiratory monitoring. Transthoracic echocardiogram revealed a good left ventricular systolic function, a mild diastolic dysfunction, abnormally distended inferior vena cava (IVC) and low systemic vascular resistance (SVR): $37.5 \text{ MPa}\cdot\text{s}/\text{m}^3$ (normal range $90\text{--}120 \text{ MPa}\cdot\text{s}/\text{m}^3$). He continued to have a low DBP, leading to an increase of dopamine dosing up to $10 \mu\text{g}/\text{kg}/\text{min}$, which was still ineffective, so norepinephrine $0.15 \mu\text{g}/\text{kg}/\text{min}$ infusion was subsequently added with immediate stabilisation of DBP. Norepinephrine was stopped after 6 hours and dopamine continued with haemodynamic stability. Further history taking and questioning about possible skin lesions led the parents to remember that he had mentioned a sacrococcygeal pain 3 days earlier. On examination, he had a slight induration in the crease just above the buttocks, painful with palpation, but skin was intact with no erythema. There were no findings on anal and perianal inspection. The sacrococcygeal ultrasound identified a poorly defined fluid collection in the proximal buttocks crease area ($50\times 20 \text{ mm}$) suggestive of pilonidal abscess. He underwent

Figure 2 Close-up of the erythematous maculopapular rash on the left lower leg, with a confluent macular lesion with central desquamation.

Figure 3 Bilateral conjunctival hyperaemia, glossitis and mild desquamation of the skin.

drainage with needle aspiration followed by saline irrigation twice. The purulent discharge was sent for culture and sensitivity, and it came positive for a penicillin G-resistant SA and *Peptostreptococcus magnus* (PM). Further microbiological analysis revealed that this SA strain produced enterotoxin (ET) type A, C and E. The initial blood and nasal swab cultures came negative.

OUTCOME AND FOLLOW-UP

He became afebrile on the second day of stay on PICU, and the rash completely disappeared on the third day. Dopamine infusion was gradually reduced and discontinued after 3 days. A repeated echocardiogram showed a normal IVC diameter, with normalisation of diastolic function and SVR. On the sixth day, he was transferred to the local hospital to ensure continuity of care. Three days after, a recrudescence of sacrococcygeal symptoms occurs, motivating another abscess drainage and irrigation. Throughout local hospital stay, he remained afebrile and haemodynamically stable. Overall, he completed 14 days of flucloxacillin and clindamycin with a complete and final resolution of symptoms. Generalised desquamation never appeared at 2 weeks or later. No recurrence occurred at 6-month follow-up in clinic. He is scheduled for pilonidal cystectomy by marsupialisation to promote faster healing and minimise the impact on quality of life.

DISCUSSION

Pilonidal sinus, derived from the Latin words pilus (hair) and nidus (nest), is an acquired condition that occurs when broken hair is driven into the skin of the natal cleft by a rolling action of the buttocks, which provokes a foreign body-type reaction with chronic inflammation.¹ Pilonidal disease occurs predominately in men (75%–80%), and there is a peak in incidence between the ages of 15 and 24 years.^{2,3} At an early stage of pilonidal disease, before the development of an abscess, only a cellulitis or folliculitis is present. The abscess is formed when a folliculitis infiltrates into the subcutaneous tissue or when an ongoing foreign-body granuloma becomes infected.⁴ Several predisposing factors of pilonidal disease have been suggested, such as hairy body, local trauma due to long periods of sitting, poor hygiene, wounded skin and positive family history.⁵ Certain anatomical features of sacrococcygeal area may promote the accumulation of hairs

in intergluteal sulcus, but this has yet to be proven.⁵ A recent study revealed that males aged 20–24 years and girls aged 10–14 years had a higher incidence of pilonidal disease, indicating that puberty, which starts earlier in girls, is an essential factor in the development of pilonidal disease.³

To the best of our knowledge, five cases of pilonidal sinus disease complicated with TSS have been reported in the literature.^{6–10}

Microbiology of pilonidal disease is usually polymicrobial with a predominance of anaerobic organisms. However, gram-positive bacteria are frequently present and are the most common agent of coinfection, resembling the association between PM and SA found in our case.^{11 12}

Both SA and *Streptococcus pyogenes* produce bacterial exotoxins, including a family of toxins known as superantigens, which are capable of causing TSS. The vast majority of TSS cases are caused by TSS toxin-1 (TSST-1). TSST-1 is accountable for 95% of the tampon use/menstrual cases and 40%–60% of the non-menstrual cases.¹³ The remaining cases of TSS are mediated through SA ETs A, B, C, D or E.¹⁴ Non-menstrual TSS is responsible for 30%–50% of all cases of TSS, one-third of which involve men.¹⁴ The classic diffuse sunburn-like rash (erythroderma) is seen with TSST-1-producing strains of SA.¹⁵ However, other staphylococcal ETs produce variable cutaneous manifestations, including bullous impetigo, urticaria, petechia or even a maculopapular rash that resembles a scarlatiniform eruption, as seen in our patient.¹⁶

In our case, we found an SA strain capable of producing ETs A, C and E. All of the three have the potential to induce TSS. The patient presented with fever, diffuse papulomacular rash, hypotension, vomiting and conjunctival hyperaemia, fulfilling five criteria of TSS,¹⁷ but only the last two are of multisystem involvement.

Even though our patient did not meet the full criteria for TSS, he still presented with classic clinical features in the context of a focal infection (pilonidal abscess). The authors hypothesise that the timely administration of fluids, antibiotics and pharmacological cardiovascular support in this patient might have interrupted the progression to TSS.

A careful physical examination is critically important, particularly in cases where symptoms are consistently neglected or omitted by young patients, as the one we describe. The diagnosis of a pilonidal sinus relies on identifying the epithelialised follicle opening which can be palpated as an area of deep induration beneath the skin.⁴ Despite the fact that the diagnosis of pilonidal disease is mainly clinical, an ultrasound may help to confirm the presence of an abscess and measure the fluid collection.¹⁸

In selecting empiric antimicrobial therapy for a pilonidal sinus abscess, clinicians should consider (1) site of infection, (2) prior knowledge of bacteria known to colonise the patient and (3) local bacterial resistance patterns. The association of flucloxacillin (β -lactamase-resistant antistaphylococcal agent) with clindamycin (effective agent against aerobic gram positive, including methicillin-resistant SA and anaerobic gram negative) given to this patient provided an ideal empirical coverage for a complicated sacrococcygeal pilonidal abscess. There is some evidence that clindamycin has the ability to inhibit toxin production by SA.¹⁴ The adequate antibiotic therapy allied to the removal of the infectious nidus (abscess drainage) allowed a quick and significant improvement in our patient. The drainage of pilonidal sinus abscess should be acknowledged as an essential part of therapy to eliminate the primary focus of the infection.^{14 18} Several surgical methods are used to treat pilonidal sinus, such as primary suture, lay open and the flap plasty techniques.¹⁹ Each

method has a certain recurrence rate and, therefore, no standard treatment approach has been determined, despite the high incidence of pilonidal disease affecting young population.¹⁹

In conclusion, the findings in this case clearly demonstrate the need for timely medical and surgical management of the pilonidal abscess, particularly in the presence of concurrent systemic illness, including full intensive care. Diagnosing a pilonidal abscess can be challenging when lesions or redness of the affected area are absent, and the patient omits the pain, as in this case. Secondary bacteraemia accompanied by toxæmia are a potential severe and life-threatening complication, leading the authors to urge clinicians to maintain a high index of suspicion for this condition.

Learning points

- ▶ Acute illness characterised by high fever, hypotension and non-pruritic generalised rash should raise suspicion of toxic shock syndrome diagnosis.
- ▶ The potentially unperceived local manifestations of a sacrococcygeal pilonidal abscess combined with the intentional omission of symptoms among teenagers emphasise the importance of a meticulous clinical history and physical examination.
- ▶ Adequate antibiotic coverage for anaerobic and gram-positive bacteria should be promptly started whenever complicated pilonidal sinus disease is suspected.
- ▶ Removing infection site with incision and drainage is mandatory in pilonidal abscesses, particularly if complicated by systemic disease.

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