CASE REPORT

In the setting of paediatric osteomyelitis do not be afraid to CAST an eye

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SUMMARY

The case commences with an innocuous right ankle injury (lateral malleolus), for which the patient, a 9-year-old boy, was placed in a temporary cast at his local hospital. Three days following this incident, the patient was diagnosed with new-onset type 1 diabetes mellitus. He was admitted to his local hospital with severe diabetic ketoacidosis appropriately treated and subsequently discharged c.1 week later clinically well. Approximately 1 week later, again he presented for a third time with a significant serosanguinous discharge from the site of the initial injury permeating through the temporary cast in place for c.2 weeks by that time. On removal of the cast, a severely invasive infection of bone and soft tissue was noted, and the patient was urgently transferred to our unit at the tertiary general hospital for further management. He underwent a series of orthopaedic and plastic surgery procedures with an eventual satisfactory outcome.

BACKGROUND

I originally chose to write this case up as there was a well-defined timeline with many different potentially interlinking problems; however, as I delved more into the case other interesting learning points emerged.

The importance of reaching a diagnosis and treating that same diagnosis and determining the contributing factors which lead to that conclusion being reached are highlighted in this case. There was evidence of a leukaemoid reaction on initial presentation with diabetic ketoacidosis (DKA), a white cell count (WCC) elevated way beyond the limits one would consider consistent with severe DKA; C-reactive protein (CRP) was also markedly elevated. However, at no point was the cast in situ on the patient's leg removed or examined—perhaps this may have prevented further events down the line or provided a focus to the hugely elevated inflammatory markers.

This case also reminds us as clinicians to try to see the bigger picture, not to get bogged down in the minutiae but perhaps to stand back and take a look at what could potentially be causing an unusual myriad and timeline of signs and symptoms.

CASE PRESENTATION

The patient, a previously healthy, fully vaccinated, 9-year-old Caucasian boy presented to his local hospital with a painful right ankle secondary to minor trauma. There was localised swelling and

reduced weight-bearing documented; no break in the skin or bruising noted. He had no intercurrent illness at the time and was otherwise afebrile and clinically well. Plain film radiograph performed at this time was unremarkable; a temporary cast was applied given a concern for a possible fracture with orthopaedic follow-up arranged.

Two days following the above visit, the patient developed new-onset polydipsia and polyuria with increasing fatigue. By the following morning, he was extremely lethargic and began vomiting. After acute review with his general practitioner, the patient was urgently referred to his local hospital owing to suspicion of new-onset type 1 diabetes mellitus (T1DM) with DKA. On arrival to the emergency department, he was severely acidotic with a pH of 6.97 and blood glucose of 42.1 mmol/L; he was transferred to the intensive care unit (ICU) where he remained for 2 days.

Following successful stabilisation of DKA, he was transitioned to subcutaneous insulin without complication. Outside of the usual battery of investigations for new-onset insulin-dependent diabetes mellitus with DKA, it is useful to note that, on initial presentation, the patient's full blood count revealed a WCC of $47.1 \times 10^9/\text{mL}$ (normal $5\text{--}12 \times 10^9/\text{mL}$) remainder of relevant laboratories listed below. Similarly, the CRP was significantly elevated at 177.1 mg/L (normal $<5 \text{ mg/L}$).

The patient was discharged home on hospital day 5 clinically well; outpatient follow-up was arranged with the paediatric diabetes service. At no time during this admission was the temporary cast addressed. Likewise, the patient did not complain of any pain or concerns with his right ankle during this admission; he remained afebrile throughout.

Six days following discharge, the patient awoke complaining of 'itchiness' under his temporary cast. In addition, his mother noticed a malodorous serosanguinous discharge dripping from the back of the cast. Of note, there was no pain or discomfort of his foot or ankle, sensation was intact and the patient denied any fevers or chills. For the third time, he returned to his local hospital where his original cast was removed. Revealed beneath was severe and extensive tissue necrosis overlying the lateral malleolus extending down to bone tissue.

At this point, the patient was emergently transferred to our services under the joint care of paediatrics and plastic surgery with additional input from orthopaedics.

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Figure 1 MRI of right ankle, deep ulceration over lateral malleolus, patchy bone marrow oedema.

INVESTIGATIONS

Initial full blood count (FBC) on presentation with DKA revealed a WCC of $47.1 \times 10^9/L$ (neutrophils $36.3 \times 10^9/L$; lymphocytes $3.8 \times 10^9/L$), Hb of 15.3 g/dL and platelet count of $535 \times 10^9/L$. CRP at this time was 177.1 mg/L (normal <5 mg/L). Venous blood gas was consistent with metabolic acidosis, normal lactate.

On presentation to our institution, WCC was $7.6 \times 10^9/L$ with an erythrocyte sedimentation rate (ESR) of 93 mm/hour (normal <10) and CRP of 19.5 mg/L. Subsequent ESR measurements were consistent with a gradual decline over the following weeks to a value of 31 mm/hour on discharge at which time the patient was transitioned to outpatient intravenous antibiotic therapy. Renal and liver function remained stable throughout.

No pathogens were isolated on routine culture from preantibiotic intraoperative tissue sampling; PCR for group A streptococcus, *Staphylococcus aureus* and *Streptococcus pneumoniae* was requested. Group A streptococcus DNA was confirmed on PCR analysis from tissue samples. Blood and urine cultures preantibiotics were sterile.

MRI of the right ankle and foot ([figure 1](#)) revealed bone marrow oedema throughout the distal fibular shaft and lateral malleolus in keeping with a diagnosis of osteomyelitis.

Figure 2 The patient's right ankle on removal of the cast, skin defect measuring 12×6 cm over lateral malleolus, exposed distal fibula with surrounding necrosis.

DIFFERENTIAL DIAGNOSIS

The differential diagnosis for this presentation includes osteomyelitis, septic arthritis, pressure necrosis due to plaster immobilisation or a venous ulcer (very unlikely in paediatric practice).

TREATMENT

On arrival to our institution, immediate exploration of his ankle area revealed extensive tissue necrosis measuring 12×6 cm over his lateral malleolar/lower fibular area; the distal fibula was exposed to the bone and growth plate area measuring 4×2 cm ([figure 2](#)). A vacuum-assisted closure (VAC) dressing was applied; tissue samples were submitted for microscopy and culture. Empiric therapy with intravenous flucloxacillin (50 mg/kg/dose, 6 hourly) and clindamycin (10 mg/kg/dose, 6 hourly) was commenced. A peripherally inserted central catheter was sited at this time. Intravenous antibiotics were continued for a total of 8 weeks, initially with flucloxacillin and clindamycin and subsequently once daily with ceftriaxone (50 mg/kg).

Figure 3 The patient's right ankle during surgical exploration after 2 weeks of VAC therapy, preskin graft application.

OUTCOME AND FOLLOW-UP

The patient was discharged following a 2 weeks of inpatient stay. He required three revisions of his wound (figure 3) until the plastic surgery service was happy to apply a skin graft obtained from his buttock area. The VAC dressing was removed and the patient was discharged home with the outpatient antibiotic team input. Weekly review was arranged at the paediatric and plastic surgery clinics. Weekly laboratory monitoring was put in place (return to baseline ESR, CRP and WCC). Given the aggressive nature of the infection, baseline evaluation of immune function was performed and noted to be unremarkable.

At last review (2 months from initial discharge), the patient's wound is healing well and he has returned fully to all normal activities. It is important, however, to remember the long-term cosmetic ramifications, despite aggressive and long-term treatment of a severe osteomyelitis such as in this patient's case (figure 4).

DISCUSSION

Given that one of the key learning points in this case is the importance of investigating significant leucocytosis in cases of DKA, it is important to determine what the literature informs us concerning the relationship between an elevated WCC and DKA.

A retrospective cohort study completed in 2001 of 247 patients over 6 years agrees that a 'diligent' search and work up for bacterial infection that may precipitate episodes of DKA is vital on initial evaluation.¹ The mean WCC of all patients (with or without infection) in this study was $17.5 \times 10^9/L$, much lower than in our patient's case. In the population with serious bacterial infection, 31% of these patients were diagnosed with new-onset T1DM. This same study determined that among all children who presented with DKA, bacterial infections were present in only 13% and viral infections in 18%.¹

A recent update on DKA determined that although patients with DKA will have leucocytosis in keeping with the level of acidosis and stress hormone release (catecholamines, cortisol) on presentation, WCC in excess of $25 \times 10^9/L$ may suggest occult infection and warrants further investigation.^{2,3}

In summary, on review of the literature available on this topic, increased WCC is common in DKA. However, for the majority of cases, this is a reflection of severity of DKA as opposed to

Figure 4 Most recent image of the patient's right ankle after full course of treatment completed, showing long-term cosmetic outcome.

underlying infection. The question in this case is did the presence of DKA in this patient distract from the elevated inflammatory and infective markers?

Another point of discussion is the importance of always removing or examining under dressings or equipment that occlude the skin. When looking at a case from an infectious disease perspective, the skin itself must be considered as an organ through which infection may gain access and become deep seated and therefore should not be omitted during physical examination.

The most common route of infection in paediatric osteomyelitis is via haematogenous spread⁴ (acute haematogenous osteomyelitis (AHO)), which involves bacteria being deposited in the metaphysis during an episode of bacteraemia via the blood vessels supplying this area of the bone. In these cases, a break in the skin is rarely encountered. There follows a nidus of infection developing in the metaphysis. Other routes such as direct inoculation (traumatic, surgical) or local invasion from a contiguous infection are less common in the paediatric population. One may postulate that, in this case, there may have been

low-grade osteomyelitis present on initial presentation (minor trauma possibly a red herring), which then advanced precipitously in the presence of DKA and elevated blood sugar. It is important to note that, as in this case, minor trauma can often be highlighted as a preceding event in patients who develop osteomyelitis. However, it is currently still unclear whether minor trauma is contributory to the overall disease process. It is not surprising that this patient remained afebrile given that fever is an inconsistent clinical presenting finding in children with AHO. The usual clinical features of haematogenous osteomyelitis in long bones have been described in keeping with the spread of the causative organism throughout the blood stream, chills, fever and general malaise, as well as the local infectious processes—pain and swelling, which should be assessed on thorough clinical examination.⁵

Learning points

- ▶ Always apply the ABCDE rule in the acute clinical setting such as during the intensive care unit admission in this case, fully expose the patient even if this involves cutting off a back slab or cast, remove bandages and perform a thorough physical examination.
- ▶ In the setting of diabetic ketoacidosis (DKA), an elevated white cell count (WCC) must always be fully investigated and relevant history obtained. DKA may be associated with an elevated WCC. However, when the value exceeds $25 \times 10^9/L$ or the patient has severe DKA on presentation, a thorough evaluation for infection should be undertaken.
- ▶ In the young child presenting with osteoarticular pain in the absence of significant trauma, consider acute haematogenous osteomyelitis as a possible diagnosis and ensure baseline inflammatory markers and careful clinical examination are performed with appropriate follow-up put in place.

In terms of imaging, MRI is very sensitive in assessing for osteomyelitis and is particularly useful in the neonate and young child, allowing the assessment of cartilaginous (including growth plate) involvement.⁶ It is worth noting that, as in this case, growth plate destruction can, not surprisingly, be linked with altered growth, including abnormal leg length or an angular deformity.⁷ It is, therefore, important that this patient have strict follow-up to determine and follow any long-term sequelae that may develop. A delay in treatment is associated with worse outcome as reported in a systemic review of the literature published in 2012.⁸

Contributors RAR completed the case report with editing advice and assistance from EM. EM was the consultant responsible for this patient during his time in our department and continues to oversee his care in the outpatient setting.

Competing interests None declared.

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