Childhood pyoderma gangrenosum: A case report

Pauline Dioussé, Ndiaga Gueye, Mariama Bammo, Thierno Abdou Aziz, Haby Dione, Amadou Mactar Gueye, Fatou Seck, Mame Thierno Dieng, Bernard Marcel Diop, Mamadou Moustapha Ka

ABSTRACT

Introduction: Pyoderma gangrenosum is a rare neutrophilic dermatosis in childhood, of unknown etiology, characterized by ulcerations.

Case Report: We report a case of a boy of 5 years, hospitalized for extensive phagedenic ulcers with a characteristic centrifugal extension for 30 months. The parents had consulted several health institutions without success. The lesions were localized on the left lower limb, to the medial aspect of the right leg and the right clavicular hollow. There was ankyloses of the right knee. The hemogram showed severe anemia. Bacteriological, mycological, viral samples were negative. Histology showed a rich infiltrate of polymorphonuclear neutrophils. No other underlying pathology was identified. The child was treated with prednisone 1 mg/kg/day in digressive manner over a period of two months with a full recovery at the end of this period. He received a blood transfusion and orthopedic surgery of his right knee, followed by rehabilitation sessions. The evolution was good leaving unsightly scars from the ulcers and a recovery of walking.

Conclusion: The peculiarity of this case lies in the extensiveness of the lesions, the intensity of the anemia and the diagnostic difficulties due to the large and chronic cutaneous ulcerations of the child in Africa. This results in delay of diagnosis and threatens functionality in life.
CASE REPORT

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Keywords: Anemia, Child, Diagnosis, Pyoderma gangrenosum, Senegal

INtRODUCtION

Pyoderma gangrenosum (PG) is an uncommon neutrophilic dermatosis in children (4% of cases) [1]. Despite the fact that the pathogenesis of PG is unknown, a variety of diseases may be associated with it, mainly ulcerative colitis, Crohn’s disease, and hematologic malignancies [1]. Isolated cases have been described in the infant and child [1, 2]. It is often generalized nature, the severity of associated anemia and diagnostic difficulties are known characteristics [3, 4]. Most children with PG have been successfully treated with oral corticosteroids. We report a case in its ulcerative form and late diagnostic.
CASE REPORT

Our case involves a boy of 5 years, received 30 months before being admitted to hospital after having consulted, without success, 6 doctors and herbalists for large ulcerations of the lower limbs. His vaccination status was up to date. The patient had no remarkable personal or familial antecedents. His developmental mile stones and general health were good. He had neither diarrhea nor arthralgia. The child was afebrile.

The dermatological examination showed extensive ulcers, well rounded as by “stroke of compass” phagedenic with a centrifugal extension, raised borders and some zones with purulent huches. Lesions were in the thigh and left leg and inside the right leg (Figure 1). We noted also an ulcer in the right clavicular hollow with extension to the thorax 7 cm in diameter on its major axis (Figure 2). There was also intense pain to touch ulcers and ankyloses of the right knee. The rest of the examination was normal.

The hemogram showed severe anemia (hemoglobin: 6 g/dl), microcytic (MCV <80 µ3), a low serum iron and siderophiline coefficient saturation and a polynuclear neutrophil leukocytosis. The reticulocyte rate was normal.

Histology showed a rich infiltrate of polymorphonuclear neutrophils in the deeper layers of the dermis.

The bacteriological samples were sterile. Bone radio knee was normal. The retroviral serology and a search of antinuclear antibodies was negative. Tuberculin test, abdominal ultrasound, arterial-venous Doppler ultrasound, digestive tube endoscopy and colonoscopy were normal.

Treatment with prednisone 1 mg/kg/d in a digressive manner over two months and local wound care allowed the beginning of the healing process (Figure 3). He was transfused with iso blood group, rhesus and orthopedic surgery of his right knee, followed by rehabilitation sessions.

The evolution was good at the end of second month with unsightly scarring due to the extensive ulcerations (Figure 4). The response of treatment was a disappearance of local pain ulcers, improvement of anemia (hemoglobin: 10 g/dl) and a recovery of walking; characteristics as summarized in Table 1.
Figure 3: Healing of the ulcers by scar tissue at the end of 15 days of prednisone, with clear margins “stroke of the compass”.

Figure 4: The unsightly scarring of ulceration of the left leg after 2 months of treatment.

Table 1: Clinical, paraclinical, therapeutic and evolutionary characteristics of patient.

<table>
<thead>
<tr>
<th>Characteristics</th>
<th>Details</th>
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<tbody>
<tr>
<td>Clinical</td>
<td>The dermatological examination showed extensive ulcers, well rounded as by “stroke of compass” phagedenic with a centrifugal extension, raised borders and some zones with purulent huches. Unsightly scarring</td>
</tr>
<tr>
<td>Paraclinical</td>
<td>The hemogram showed a poly nuclear neutrophil leukocytosis. Histology showed a rich infiltrate of polymorphonuclear neutrophils in the deeper layers of the dermis. The bacteriological samples were sterile</td>
</tr>
<tr>
<td>Therapeutic</td>
<td>Treatment with prednisone 1 mg/kg/d in a digressive manner over two months and local wound care allowed the beginning of the healing process.</td>
</tr>
<tr>
<td>Evolutionary</td>
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</tr>
</tbody>
</table>

In our patient, we note that difficulties of access to dermatologic services coupled with certain cultural beliefs caused the patient to seek alternative therapies impacting prognosis of the condition. It is this interval before initiating an adequate treatment, which is at the origin of the extensive lesions and stiffness in the left knee making mention ankylosis. Radiographs of the knee showed a bone integrity moving towards ankylosis soft tissue. There was no septic arthritis objectified or acne (as part of a PAPA syndrome: arthrite syndrome purulent-acne-pyoderma gangrenosum) or osteomyelitis (as part of a syndrome Majeed: neutrophilic dermatosis-anemia-osteomyelitis). To behave was surgery of releasing the posterior soft parts then a full knee extension followed by a fasciocutaneous plasty recovery then establishment of a plaster alternating with early physiotherapy.

Neutrophilic diseases may be associated with chronic inflammatory diseases of the digestive tract, neoplasia, and dysglobulinemias [1, 7]. The various tests carried out to exclude one of these entities were negative. However, close and prolonged monitoring is necessary because the PG may precede one of these conditions. The etiopathogenesis of the PG is still poorly known. The prognosis was good on corticosteroid therapy. In the case of cortico-resistance, some authors recommend immunosuppressant. Currently, the anti-TNFα is a promising treatment of refractory PG [8].

**CONCLUSION**

We have reported our observation of pyoderma gangrenosum in a child in its ulcerative form. Its uniqueness lies on the extensiveness of the lesions, the
diagnostic difficulties due to the intensity of the anemia and the chronic extensive cutaneous lesions of the child, all lead to delay in diagnosis and consequently impacting the functionality of and or life itself.

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Author Contributions
Pauline Dioussé – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Ndiaga Gueye – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Mariama Bammo – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Thierno Abdou Aziz Diallo – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
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Guarantor
The corresponding author is the guarantor of submission.

Conflict of Interest
Authors declare no conflict of interest.

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