PERIPHERAL GANGRENE IN AFRICAN CHILDREN : A CLINICAL REPORT OF TWELVE CASES

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Clinical observations of 12 cases of peripheral gangrene in children are reported. All patients presented with ischemia of one or more limbs without any history of trauma, vascular injury or snake bite. Prior to their admission, all these children had received some form of traditional "African" therapy. By means of exclusion severe vasospasm secondary to the traditional treatment was considered the causative factor of the peripheral gangrene in all the children.

Surgical exploration of the arteries with a Fogarty embolectomy catheter in two patients and medical treatment in six patients was attempted. The overall results were disappointing, with most resulting in incapacitating amputations.

Keywords : peripheral gangrene ; vasospasm ; traditional "African" therapy.

Mots-clés : gangrène périphérique ; vasospasme ; médecine africaine traditionnelle.

INTRODUCTION

Peripheral gangrene in children is uncommon. The exact etiology is often unknown. A number of reports have demonstrated that sudden changes in the blood clotting mechanism can occur in patients with bacterial septicemia.

Miller (7) described a condition "tropical coagulopathic ischemia" when peripheral gangrene was seen secondary to a gross disturbance of the balance between coagulation and fibrinolysis. This can be seen in chronic ill health such as severe malnutrition, chronic diarrhea or pulmonary tuberculosis.

Gangrenous extremities were already described in ancient and medieval times with epidemics of ergotism, caused by the ingestion of bread or cereal made from rye contaminated by the fungus *Claviceps purpurea* or ergot. The term St. Antony's fire was used to describe the gangrenous extremities, which were blackened like charcoal and said to be consumed by the "holy fire", with relief obtained at the shrine of St. Anthony. The last major reported epidemic of ergotism occurred in Ethiopia in 1977 and 1978.

The condition of "idiopathic peripheral gangrene" as described in this report was seen in 11 otherwise healthy children. In Zimbabwe it is common practice that patients seek advice from a N'anga or traditional medical practitioner. A N'anga is not only a minister of religion but also a diagnostician and healer. Diseases are treated by divination, oral herb preparations, enemas, scarification or inhalations. Inhalations are commonly used for respiratory disorders such as asthma, to drive away bad spirits. One way is to inhale under a towel or blanket, smoke from herbs made into a form of powder (Nbanda), which is added to boiling water. This treatment with traditional therapy was considered the causative factor of peripheral gangrene in all of the cases.

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PATIENTS AND METHODS

Twelve children with ischemic gangrene of the extremities were studied. All of the patients were black, seven were female and five male. The mean age was 16 months with a range of 2 to 60 months. There was in no instance any history of trauma, vascular injury or snake bite. The children were not affected by sickle cell anemia. None had cyanotic disease or any clinical sign of thrombophlebitis. All the patients had received, prior to their admission, some form of "traditional African therapy", mostly by inhalation.

Seven out of 12 patients were initially seen in the Cardiovascular or in the Neurosurgical Department at the Mpilo Hospital in Bulawayo. Over a period of 22 months (Feb. 1985 to Nov. 1986) about 9500 children not older than ten years of age were admitted to this central hospital, which is the main referral center of Matabeleland in Zimbabwe. Out of this total of 9500 patients there were 7 patients with peripheral gangrene. Of these seven patients, three children (cases 2, 4 and 5) were admitted with either respiratory or gastro-intestinal symptoms, and two children (cases 1 and 7) with neurological symptoms (stupor, apathy). Two children (cases 3 and 6) presented with general malaise. On admission, the child in case 3 was dehydrated. The patient in case 6 had a peripheral infected focus secondary to chronic osteomyelitis of the femur. Roentgenograms of the chest were taken in cases 2, 4 and 5. A lumbar puncture was done in cases 1 and 7. No coagulation tests were carried out in any of these seven patients. The main medical treatment consisted of anticoagulants and/or vasodilatators, as shown in table I. Surgical exploration of the arteries with a Fogarty embolectomy catheter was performed in cases 2 and 7.

Five out of 12 patients were seen by the senior author in the Orthopedic Department of the two central hospitals of the University of Zimbabwe in Harare over a period of 28 months (Jan. 1988 to May 1990). These five children presented with well-established ischemia of one or more extremities (fig. 1). Coagulation tests were done in two of the patients (cases 8 and 9). In none of these five patients was medical treatment or surgical exploration of the arteries performed.

RESULTS

The results are as summarized in table I.

In the seven patients seen initially in the cardiovascular or neurosurgical department, clinical signs of ischemia (swelling, bluish discoloration and blisters) were first noted at least 24 hours after admission. The ischemia was progressive in all of the patients. The delay before specific management of ischemia was administered ranged from 10 to 48 hours, except for the child in case 3 who died some hours after onset of the ischemia.

Fig. 1. — Clinical picture of a 6 month-old boy (case 8) with peripheral gangrene of both feet and left hand.

The roentgenograms of the chest taken in cases 2, 4 and 5 were normal. The results of a lumbar puncture, done in two patients (cases 1 and 7) were negative.

In the two patients (cases 2 and 7) who underwent surgical exploration of the arteries with a Fogarty embolectomy catheter, no thrombi were present, but the arteries were found to be in extreme vasospasm.

Of these seven patients, two died and five were treated surgically by an amputation at the most distal level.

In the five patients seen in the orthopedics department no medical treatment was administered. Coagulation tests done in two patients (cases 8 and 9) were normal. Surgical treatment was limited to amputations at the level of ischemia.

The overall results of the surgical and medical treatment were very disappointing. Two out of 12 patients died. Ten patients survived, but in most cases extensive amputations were necessary, as shown in table I.

Case	Age (months)	Sex	Localization interventional treatment	Medical treatment	Surgical treatment
1	2	F	Both feet	Heparin	Autoamputation of the 2nd and 3rd R toes & 2nd L toe
2	3	F	L foot	Heparin Surgical exploration Nitroprusside	Transmetatarsal amputation
3	7	F	4th right toe	—	Died 4 hours after onset of ischemia
4	7	M	R foot L toes	Nitroprusside	Died
5	10	F	Both feet & legs	Heparin Phenoxybenzamine	Amputation of toes L foot
6	48	F	L foot & leg	Dextran	Above-knee amputation
7	60	M	Both feet & legs both hands	Surgical exploration Nitroprusside Dextran	Bilateral below-knee amputation Amputation of 2, 3 and 5 th L fingers
8	6	M	Both feet L hand	—	R Transmetatarsal and L Lisfranc amputation. Transmetacarpal amputation L hand
9	12	M	Both feet & hands	—	Lisfranc amputation L & R foot. Amputation of 4 fingers L hand
10	11	F	Both feet & legs	—	Bilateral below-knee amputation
11	15	M	Both feet	—	Transmetatarsal L, amputation of all toes R
12	10	F	Both feet	—	Bilateral transmetatarsal amputation

DISCUSSION

Although uncommon, there are several reports of peripheral gangrene of unknown etiology in African patients. However, apart from the article of Sinzobahamyva and Kalangu (8), there are to our knowledge no other publications describing this condition in Zimbabwe.

Peripheral gangrene has been observed in chronic ill health such as severe malnutrition, chronic diarrhea or pulmonary tuberculosis, as a result of gross disturbance of the balance between coagulation and fibrinolysis (2, 6, 7). This condition was described by Miller as "tropical coagulopathic ischemia (7).

Disorders of blood coagulation with peripheral gangrene can be seen as well in a variety of bacterial and viral infections as in tuberculosis, measles or in chickenpox. With improved antibiotic and parenteral therapy this condition has become a rarity in the Western world.

Peripheral gangrene is a well-known but uncommon complication of alpha-receptor stimulating drugs. Several authors suggested abnormal vasospasm, secondary to traditional therapy with herbs containing ergot-like substances as a possible cause of idiopathic gangrene in African adults and children (2, 6, 7, 8). Bhana and Haddeley (2) studied a pregnant woman who had been treated with a local preparation "emumbwa". The

authors attributed the cause of the gangrene to this herbal prescription. Colbert and Lysaght (4) reported gangrene in pregnancy caused by root alkaloids in a Nigerian female. Ergot-like alkaloids were extracted from a sample of the preparation.

Although specific vasospastic substances could not be identified in our cases, there was enough evidence of their presence in the two children who had surgical arterial exploration and a strong suspicion in all the other cases. By means of exclusion severe vasospasm secondary to traditional therapy was considered the causative factor in all of the cases of peripheral gangrene presented in this study. Considering the similarity to gangrenous extremities seen from ergotism, we assume that a substance similar to ergot was used. It is known that ergotism can be caused by the ingestion of bread or cereal made from rye contaminated by the fungus *Claviceps purpurea*, or ergot. In our cases, unfortunately, it was not possible to detect the exact nature of the herb used. It appears that a N'anga may prescribe a certain herb for a particular disease, while the same herb is given by another N'anga for a very different complaint. Moreover N'anga's keep their treatment secret from outsiders, and usually the patients' history is of very little help.

The appreciation of the role of vasospasm is important. Medical treatment should include vasodilators. The administration of anticoagulants alone is not appropriate since vasospasm seems to be the causative factor. In this series nitroprusside, a potent direct smooth muscle vasodilatator, was used in three cases. As an alternative to nitroprusside, prazosin hydrochloride (which reduces the peripheral vascular resistance by alpha adrenergic blockade) can be used. The administration of phenoxybenzamine (used in one case in this series), tolazoline, procainamide and reserpine have varying effects and the benefit of these drugs is doubtful.

In addition, treatment with heparin sodium and low-molecular-weight dextran may be considered. Heparin sodium may reduce thrombus formation where necrosis has occurred and may prevent endothelial changes. Low-molecular-weight dextran decreases the viscosity and sludging of blood in small vessels and may be helpful.

The use of mechanical intra-arterial balloon dilatation has been reported in the treatment of ergotamine-induced peripheral vasospasm (9). This technique was not used in this series.

The results of the medical treatment depend on the severity of the associated infection or the underlying intoxication, as well as on the delay between onset of the symptoms and the anti-ischemic therapy. In peripheral gangrene, as described in this report, prompt and appropriate medical treatment is of utmost importance and may reduce the mortality and limb loss.

Since tissue regeneration is high in children, surgery should be undertaken, as limb sparing as possible. However, the results are very disappointing with frequent deaths ; most result in incapacitating amputations.

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SAMENVATTING

J. F. NOYEZ, N. SINZOBAHAMVYA, K. KALANGU. Perifeer gangreen bij kinderen : bespreking van twaalf gevallen.

Twaalf gevallen met perifeer gangreen werden geevalueerd. Alle patiënten vertoonden ischaemie ter hoogte van één of meerdere ledematen. Er waren geen tekenen van vasculaire insufficientie, uitlokkend trauma of slangenbeet in de voorgeschiedenis. Voor opname in het ziekenhuis werden alle kinderen behandeld door één of andere traditionele Afrikaanse geneesmethode.
Vasospasm secondair aan deze traditionele behandeling werd weerhouden als uitlokkende oorzaak van perifeer gangreen. Chirurgische exploratie van de arteries met een Fogarty embolectomie catheder bij twee patiënten en een medische behandeling bij zes patiënten werd uitgevoerd.
De resultaten waren zeer ontgoochelend, meestal met invaliderende amputaties als eindresultaat.

RÉSUMÉ

J. F. NOYEZ, N. SINZOBAHAMVYA, K. KALANGU. Gangrène périphérique chez l'enfant africain : à propos de douze cas.

Douze enfants présentant une gangrène périphérique furent évalués. Tous présentèrent des signes d'ischémie au niveau d'un ou de plusieurs membres. Il n'y avait pas de signes d'insuffisance vasculaire, pas d'antécédent traumatique ni de morsures de serpent. Avant l'admission les enfants avaient été traités par une médecine traditionnelle africaine.
Un vasospasme secondaire à ce traitement traditionnel fut considéré comme l'agent de la gangrène périphérique. L'exploration chirurgicale des artères avec embolectomie à l'aide d'une sonde de Fogarty fut effectuée chez deux patients. Chez six autres patients, on a eu recours à un traitement médical.
Les résultats furent décevants, avec pour la plupart, des amputations invalidantes comme résultat final.