

Case Report

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Gangrene of the Left Forearm Following Septicaemia in a Nigerian Child With Tetralogy of Fallot: A Case Report

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ABSTRACT

Tetralogy of Fallot (TOF) carries a higher risk for infective endocarditis (IE) due to associate multiple cardiac lesions. The incidence of IE in operated TOF patients either by corrective or palliative surgery is high (around 18%), however small (around 4%) in non-corrected TOF. Most reported cases of infective endocarditis complicating TOF have been in adults, reports of central embolization complicating infective endocarditis is also rare in African literatures, especially in children with TOF. We therefore report a case of gangrene of the left forearm from central embolization in a 14-month child with TOF who had septicaemia with infective endocarditis and acute renal failure.

KEYWORDS: Infective endocarditis; Gangrene; Septicemia; Acute renal failure; Tetralogy of fallot (TOF).

INTRODUCTION

The incidence of infective endocarditis (IE) in congenital heart disease (CHD) is increasing especially in adults. This may be due to advancement in cardiac surgery and catheter intervention that led to increase in the number of patients with complex congenital heart diseases surviving to adulthood.¹ Infective endocarditis risk in CHD varies from 1% to 9%.^{2,3} Tetralogy of fallot (TOF) carries a higher risk for IE due to associate multiple cardiac lesions.⁴ The incidence of IE in operated TOF patients either by corrective or palliative surgery is high (around 18%), however small (around 4%) in non-corrected TOF.^{3,5} Most reported cases of infective endocarditis complicating TOF have been in older children and adults^{6,7} reports of central embolization is also rare in African literatures, especially in children with TOF. We therefore report a case of gangrene of the left forearm from central embolization in a 14-month child with TOF who had septicaemia with infective endocarditis and acute renal failure.

CASE REPORT

A 14-month-old female toddler presented at our centre with 2 weeks of fever and 3 days of loss of consciousness. Two days before presentation, gangrene of the fingers and left forearm was noticed, the following day, urinary output was observed to be reduced. On examination, she was unconscious and febrile (39.5 °C): She had features of down syndrome, anasarca and low oxygen saturation (94%) on 2 L/min of oxygen *via* nasal prongs. There was dark discoloration of the left upper limb extending from the fingers to distal two-thirds of the forearm with hyperemic patches on the medial aspect of the elbow. The fingers were cold, shrunken, in fixed flexion deformity and absent radial and brachial pulses. Doppler studies showed that the brachial, radial and ulnar arteries were not audible.

She presented at the referring centre a week earlier with three-month history of recurrent fast and difficult breathing and a week history of fever. She was observed to be centrally cyanosed. Investigations in the hospital showed a packed cell volume of 48%, total white cell count (WBC) of 29,800 cells/mm³ with relative neutrophilia and a platelet count of 210,000 cells/mm³. Blood culture yielded β hemolytic *Streptococcus* species sensitive to ciprofloxacin, erythromycin and pefloxacin. A diagnosis of TOF with septicemia in a patient with Down syndrome was made. She was hypoxic, hypercarbic and acidotic with PO₂ of 39 mmHg; PCO₂ of 28.6 mmHg; pH of 7.15, HCO₃ of 10 mEq/L, creatinine of 0.6 mg/dl.

At the referring centre, acidosis was corrected and she was nursed on intranasal oxygen. Blood transfusion and intravenous fluids were given and she was placed on antibiotics (ciprofloxacin, vancomycin and oral erythromycin). Ranitidine and propranolol were also administered. She had repeated episodes of hypercyanotic spells and lapsed into unconsciousness on the 4th day of admission in the referring hospital. This was followed by gangrene of the left upper limb noticed two days later. Oliguria with a urine output of 0.4 ml/kg/hr and edema were noticed on the day of referral and she was commenced on intravenous dopamine infusion at the rate of 22 μ g/kg/hr which was discontinued at the point of referral.

At presentation echocardiography confirmed TOF with biventricular dysfunction. The packed cell volume was now 55.9%, with leucocytosis (total white cell count of 19,600 cells/mm³) and thrombocytopenia (platelet count of 14,000 cells/mm³). Antibiotic treatment was changed to intravenous cefotaxime, ampicillin and metronidazole. The parents declined the offer of limb amputation and other investigations including cranial computerized tomography which could not be done before the patient died.

DISCUSSION

Tetralogy of Fallot is a cyanotic congenital heart disease char-

acterized by chronic hypoxaemia.⁷ The diagnosis in the patient being reported was heralded by features suggestive of hypercyanotic spells, a complication of TOF.⁸ Limb gangrene is a known complication of TOF following classical Blalock-Tausig shunt.⁸ The patient in this report however did not undergo this procedure. As far as the authors are aware, digital gangrene is not a common documented complication in TOF patients before surgical intervention.

The cause of the left forearm and finger gangrene in the patient is not immediately clear (Figure 1).

A possible cause is central embolization involving the left brachial artery from infective endocarditis complicating the *streptococcus septicaemia*. TOF carries a higher risk for IE due to associate multiple cardiac lesions,⁴ and embolic events have been reported to occur in 22-44% of patients with IE,^{9,10} although transthoracic echocardiography did not reveal vegetations in this patient, a more sensitive trans oesophageal echocardiography which was not available in the centre) might have helped in confirming this diagnosis.^{11,12} Another possible cause may be disseminated intravascular coagulopathies (DIC) complicating the septicemia. The possibility of DIC is suggested by thrombocytopenia observed in our centre compared after a normal platelet count at the referring centre. It could have resulted from immune-complex damage to the vascular endothelium leading to stimulation of a cascade of reactions responsible for haemostasis. The clots formed are dissolved through the fibrinolytic pathway inducing more clot formation with eventual consumption of clotting factors. There are three possible sequels to this derangement. First, the vasculitis may result in symmetrical diffuse hemorrhages in the form of petechiae and purpura. Secondly, the dissolved clots may embolise and impact on distal arterioles causing ischaemic necrosis and gangrene. Thirdly consumption coagulopathies may eventually lead to uncontrollable bleeding from various sites¹³ but this could not be confirmed because parents declined coagulation profile assay. In any case, gangrene from DIC may be expected to be symmetrical.¹⁴⁻¹⁷ Adeodu and Senbanjo¹⁷ documented digital gangrene in



Figure 1: Ischaemic gangrene involving the left forearm.

a four and half year old girl who had septicemia. The gangrene in that case was symmetrical and there was no suggestion of congenital heart disease. Bugaje et al¹⁸ in 2009 also reported an unusual case of lower limb gangrene in a pubertal Nigerian boy following *Salmonella typhi septicemia* although the described case was reported not to have clinical or laboratory evidence suggestive of DIC or coagulopathies. Other authors^{15,16} also described symmetrical peripheral gangrene in adult patients with septicemia, DIC in our patient could also have been the result of chronic hypoxia. Cellular hypoxia predisposes the tissues to reperfusion injury leading to local vasoconstriction, thrombosis, regional perfusion, release of superoxide radicals and direct cellular damage, subsequent activation of neutrophils and release of pro-inflammatory cytokines like tumor necrosis factor (TNF), interleukin-1 (IL-1), and platelet activating factor result in cellular injury, organ dysfunction and failure and death.¹⁹

Another plausible reason for the digital gangrene may be secondary to polycythaemia,²⁰ which might have followed the blood transfusion. Polycythaemia causes gangrene due most probably to impaired perfusion resulting from hyperviscosity.²¹ However; the packed cell volume at presentation was only 55%.

The possibility of dopamine associated gangrene cannot be ruled out. In low doses (2-5 µg/kg/min), dopamine brings about vasodilatation in the renal and mesenteric vascular beds. In moderate doses (10-20 µg/kg/min), it enhances cardiac contractility but higher doses (20-50 µg/kg/min) may cause vasoconstriction.²¹ Consequently, peripheral ischemia and gangrene are not unexpected following the use of large doses of dopamine. Gangrene has only rarely been reported with dopamine infusion rates in the range of 1.5-10 µg/kg/min.²¹ Gangrene complicating low-dose dopamine therapy suggests either an idiosyncratic response to the drug or a multifactorial cause of the ischemia and necrosis.²¹ It is important to note that dopamine must be infused through a central vein²² and the patient monitored in an intensive care unit where patient should be carefully monitored during dopamine therapy for decreased circulation to the extremities. Our patient received a high dose of dopamine, and there was no indication that this was under an intensive care monitoring nor through a central vein. It is however difficult to attribute the limb gangrene in our patient to dopamine since the gangrene had been noticed before commencement of this inotrope. It is arguable that the high dose given to our patient and might have contributed to or worsened the ischemia and necrosis in this patient.²¹

Ischemia or necrosis from dopamine use may be corrected by decreasing the rate of infusion or discontinuing dopamine. However, the potential benefits of continuing dopamine should be weighed against the possible risk of necrosis.²¹ To reverse ischemia due to dopamine, 10 mg of chlorpromazine given *via* intravenous route followed by chlorpromazine infusion of 0.6 mg/minute has been used. Some clinicians recommend IV administration of 5-10 mg of phentolamine (an alpha-blocking agent) if discoloration of extremities occur.²¹ Topical glyceryl trinitrate ointment is known to improve capillary blood flow in

patients with dopamine-induced digital ischemia.²¹ It is important to note that the use of dopamine infusion should be undertaken with extreme caution and only in centres where proper monitoring can be done.²¹ Inadequate funding, dearth of manpower as well as insufficient support services from the laboratories among others makes this a daunting task in developing countries.^{22,23}

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CONFLICTS OF INTEREST

The authors declare that they have no conflicts of interest.

CONSENT

The authors have received written permission for the publication of this case detail.

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