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## **Original Research Article**

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# Lower extremity gangrene in children from traditional bone setters care: an avoidable cause of limb loss

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#### **ABSTRACT**

**Background:** Worldwide, lower extremity gangrene in children is rare. In orthopaedic practice in developing countries, a frequent cause of extremity gangrene is the practice of traditional bone setting. Aim of the study was to document lower extremity gangrene in children resulting from the care of traditional bone setters' as seen in a tertiary health institution.

**Methods:** A prospective study of children presenting with lower extremity gangrene from the care of traditional bone setters at the University of Port Harcourt Teaching Hospital from January 2006 to December 2018. Data obtained included the patients socio-demographics and relevant information related to the diagnosis. Data was analysed with SPSS version 20.

**Results:** Eighteen children with lower extremity gangrene were seen. Their ages ranged from 10 days to 132 months. There was a female preponderance. Their parents mostly had primary and secondary levels of education and majority were married. The most common diagnosis was tibiofibular fractures followed by congenital clubfoot. Trauma-related injuries were mostly from falls. Duration before presentation ranged from 7 to 28 days. Majority had massage, splinting and bandaging from the bone setter. Most common associated complication at presentation was sepsis. All had provisional amputation. Most common amputation was below knee. Most had stump refashioning within 15 days. One had split skin grafting. There was one mortality. Duration of hospital stay ranged from 3 to 65 days.

**Conclusions:** Children at any age can develop lower extremity gangrene from traditional bone setters' care. This is an avoidable and preventable sequelae of care. Parental health education is necessary to discourage their patronizing traditional bone setters.

Keywords: Children, Gangrene, Lower extremity, Traditional bone setters

#### **INTRODUCTION**

Worldwide, lower extremity gangrene in children is rare. 1-3 Lower extremity gangrene in children has been associated with birth asphyxia, Rhesus disease, respiratory distress, severe congenital anomalies, maternal diabetes, invasive vascular access, dissecting femoral artery intramural haematoma, in utero arterial thrombosis, intravenous hyperosmolar infusion,

gastroenteritis, bacterial and viral infections as well as parasitic infestations, symmetrical peripheral gangrene and tropical idiopathic lower limb gangrene. <sup>1-9</sup> In orthopaedic practice in developing countries, a frequent cause of extremity gangrene is the practice of traditional bone setting. <sup>9-12</sup> Traditional bone setting is widespread in developing nations, existed long before the arrival of orthodox medicine and the practitioners enjoy wide patronage from the community. <sup>13-18</sup> The practitioners are

uneducated or barely educated and they rely more on experience and spiritual intuition. <sup>14-16</sup> The practice is preserved in the family and training is by apprenticeship. <sup>14-17</sup> Traditional bone setters treat fractures and other orthopaedic problems. <sup>16</sup> It has been estimated that up to 60-70% of trauma is treated by traditional bone setters. <sup>13,16</sup> The practice is not without complications, one of which is extremity gangrene.

The aim of this study was to document lower extremity gangrene in children resulting from the care of traditional bone setters as seen in a tertiary health institution in southern Nigeria.

#### **METHODS**

This was a prospective study of consecutive children aged 16 years or less presenting with lower extremity gangrene from the care of traditional bone setters at the University of Port Harcourt Teaching Hospital, Port Harcourt, Rivers State, Nigeria from January 2006 to December 2018.

The patients with traditional bone setters' gangrene of the lower extremity were assessed clinically, admitted and resuscitated with intravenous fluids and given broad spectrum antibiotics and tetanus prophylaxis as well as anti-tetanus regimen if tetanus was present. Relevant investigations included full blood count, serum electrolytes, urea and creatinine levels and grouping and cross-matching of blood. Plain radiographs of the affected extremities were reviewed for those who presented with them. Those who required blood transfusion were given. All the patients had provisional amputations and stump refashioning or other relevant procedures subsequently.

The inclusion criteria were those aged 16 years or less, presenting with lower extremity gangrene from the care of traditional bone setters. Excluded were patients whose lower extremity gangrene were due to crush injury from trauma and those who were older than 16 years.

Data was collected through a structured proforma and included their age, sex, parents highest educational level, parents marital status, primary diagnosis, cause of injury, duration before presentation, traditional bone setters treatment, duration of traditional bone setters treatment, mode of traditional bone setters care, associated complications at presentation, amputation level, duration before stump refashioning, other additional procedures carried out, duration of hospital stay, mortality and duration of follow up.

Data was analysed using Statistical Package for Social Sciences (SPSS) version 20 (IBM Inc., Armonk, NY, USA). Mean, standard deviation and median were used for descriptive statistics while categorical variables were expressed in absolute frequencies. Spearman's correlation was used to determine association between

sociodemographic characteristics and the amputation level. A p value of  $\leq 0.05$  was considered significant.

Ethical approval was obtained from the Research and Ethics Committee of the University of Port Harcourt Teaching Hospital.

#### **RESULTS**

During the period, 18 children who were 16 years or less were seen with traditional bone setter's gangrene of the lower extremity. This constituted 3.0% of the total admissions into the children orthopaedic ward and 0.4% of the total admissions into the children surgical wards. Their ages ranged from 10 days to 132 months with a mean of 52.60±44.55 months. There were six infants and two of these were neonates; the rest were older. There were 10 females and eight males. The parents' highest level of education were mostly primary (50.0%) and secondary (44.4%). There was none that had tertiary level of education. Most of the parents were married (72.2%) although 16.7% were separated (Table 1).

Table 1: Socio-demographic characteristics of patients.

Variables	Frequency (n=18)	Percentage	
Age			
<25 months	6	33.3	
25-48 months	3	16.7	
49-72 months	3	16.7	
73-96 months	3	16.7	
>97 months	3	16.7	
Mean (SD), Median	52.60(±44.55) 54.00		
Range	10days - 132months		
Sex			
Female	10	55.6	
Male	8	44.4	
Parents highest level of education			
None	1	5.6	
Primary	9	50.0	
Secondary	8	44.4	
Parents marital status			
Divorced	2 11.1		
Married	13	72.2	
Separated	3	16.7	

The gangrene affected the left side in seven children and in 11 it was on the right. In half of the patients, the diagnoses were tibiofibular fractures and congenital clubfoot in 33.3% (Table 2). For the tibiofibular fractures there were five males and four females and for clubfoot there were five females and one male (Table 3). In those whose primary injuries were the result of fractures, the most frequent cause was fall in six patients followed by

road traffic crashes (5). More males were affected (Table 4). The duration before presentation ranged from seven to 28 days with mean of  $18.50\pm6.91$  days. In the majority (77.8%) the traditional bone setters treatment consisted of

massage with herbal concoctions, splinting and bandaging. The duration of treatment with the traditional bone setter ranged from two to 28 days with a mean of  $16.11\pm6.74$  days (Table 2).

Table 2: Clinical characteristics of patients.

Variables		Frequency (n=18)	Percentage
	Tibiofibular fracture	9	50.0
Primary diagnosis	Congenital clubfoot	6	33.3
	Femoral fracture	2	11.1
	Ankle fracture	1	5.6
	RTC	5	41.7
Cause of injury (n=12)	Fall	6	50.0
J. J.	Sporting injury	1	8.3
	1-7 days	1	5.6
Duration before	8-14 days	8	44.4
presentation	15-21 days	4	22.2
•	22-28 days	5	27.8
Mean (±SD), Median		18.50 (±6.91), 16.00	
Range		7-28 days	
	Massage + Splinting + Bandaging	14	77.8
TBS Treatment	Massage + Splinting + Bandaging + Incision	4	22.2
	1-7 days	1	5.6
Duration of TBS	8-14 days	11	61.1
Treatment	15-21 days	3	16.7
	22-28 days	3	16.7
Mean (±SD), Median	•	16.11 (±6.74), 14.00	
Range		2-28 days	
Associated complication (n=	=10)	•	
Sepsis		9	90.0
Septicaemia / tetanus		1	10.0
	Mid foot	1	5.6
Amputation level	Below knee	14	77.8
	Above knee	3	16.7
	<9 days	3	18.8
Duration before stump	9-15 days	8	50.0
refashioning (n=16)	16-22 days	3	18.8
	>22 days	2	12.5
Mean (±SD), Median		15.63 (±8.54), 13.00	
Range		8-38 days	
	<22 days	1	5.6
	22-28 days	7	38.9
Duration of hospital stay	29-35 days	3	16.7
	36-42 days	3	16.7
	>42 days	4	22.2
Mean (±SD), Median		33.61 (±14.54), 30.00	
Range		3-65 days	
	1-6 months	8	47.1
Follow up duration (n=17)	7-12 months	6	35.3
	>12 months	3	17.6
Mean (±SD), Median		8.47 (±6.97), 7.00	Mean (±SD), Median
Range		2-26 months	

RTC = Road traffic crash, SD = Standard deviation, TBS = Traditional bone setter

Table 3: Cross tabulation of primary diagnosis and sex of the patients.

	Sex			
Diagnosis	Male	Female	Total	Percentage
Tibiofibular fracture	5	4	9	50.0
Congenital clubfoot	1	5	6	33.3
Femoral fracture	1	1	2	11.1
Ankle fracture	1	0	1	5.6
Grand total	8	10	19	100

Table 4: Cross tabulation of cause of injury and sex of the patient.

	Sex			
Cause of injury	Male	Female	Total	Percentage
RTC	3	2	5	41.7
Fall	3	3	6	50.0
Sporting injury	1	0	1	8.3
Grand Total	7	5	12	100

Table 5: Relationship between Socio-demographic characteristics and level of amputation.

Variables	Spearman's rho (r)	Grading	p- value
Age at presentation	0.546	Moderate	0.019
Sex	0.015	Very weak	0.953
Educational level of parent	0.261	Weak	0.296
Marital status of parent	-0.395	Weak	0.104

The patients were treated as out-patients by the traditional bone setters. Sepsis was noticed at presentation in most of the children although one had septicaemia and generalized tetanus. The patients all had provisional amputation which in the majority was below knee (77.8%). The amputation stumps were refashioned eight to 38 days after (Table 2). The refashioned stump healed without complication. One patient had had split skin grafting. Their duration of hospital stay ranged from three to 65 days with a mean of 33.61±14.54 days (Table 2). There was one mortality (5.6%). This was a five-year old female who had femoral fracture, septicaemia and generalized tetanus. The duration of follow up ranged from two to 26 months with the most frequent category being those who were followed up for up to six months (47.1%). One of the mothers was a psychotic patient.



Figure 1: Right lower extremity gangrene in a 5-year old following care for right femoral fracture by a traditional bone setter.



Figure 2: Left foot gangrene in a neonate following care for left congenital clubfoot by a traditional bone setter and the left foot stump following amputation of the gangrenous part. (A and B) are the dorsal and plantar views respectively of the foot at the initial presentation of the patient. (C and D) show further demarcation of the gangrenous foot (plantar view and medial/plantar view respectively). (E) The left foot stump following amputation of the gangrenous part.

Table 5 shows the relationship between the sociodemographic characteristics and the amputation level. There was a significant positive correlation between the patients' age and the amputation level (p=0.019). There was no correlation between the sex, parents' educational level and marital status with the level of amputation.

Figure 1 is a clinical photograph showing right lower extremity gangrene in a 5-year-old female from the care for right femoral fracture by a traditional bone setter. The same child also had generalized tetanus.

Figure 2 (A to E) are the clinical photographs of a neonate with left foot gangrene following care for left congenital clubfoot by a traditional bone setter and that after amputation of the gangrenous part. Figures 2A and 2B are the dorsal and plantar views respectively of the foot at the initial presentation of the patient. Figures 2C and 2D shows further demarcation of the gangrene. Figure 2E shows the stump of the left foot following amputation of the gangrenous part.

#### **DISCUSSION**

This study has shown that children at any age can develop lower extremity gangrene from traditional bone setting, females being affected more with the primary diagnoses being mostly tibiofibular fractures and congenital clubfoot. The traditional bone setters' treatment consisted mainly of massage, splinting and bandaging and sepsis was a major complication at presentation with the amputation being carried out in two stages with good outcome.

In the literature, majority of the publications on bone setters' gangrene of the lower extremity were discussed with amputations generally and not necessarily as a separate entity. Hence, the characteristics of lower extremity gangrene were subsumed in the discussions on amputation.

In the present study, six of the patients (33.3%) were infants and two of these were neonates while the rest were older. Children in this age group are particularly at risk as they may not be able to complain except by crying.

There was a female preponderance in the present series. Generally, males are more affected in trauma than females and would ordinarily have been predominant. <sup>19,20</sup> However, the present series included both traumatic and non-traumatic conditions. There were more females among those who had congenital clubfoot and this overshadowed the slight male predominance in the trauma-related conditions. Hence, the overall female preponderance.

Traditional bone setters engage in the treatment of traumatic and non-traumatic orthopaedic disorders. <sup>13,16</sup> In the present study, they were involved in the management

of fractures which constituted majority of the diagnoses as well as the management of congenital clubfoot. Their meddling with these resulted in the lower extremity gangrene. More pathetic is the fact that the clubfoot patients were infants. Clubfoot care has been revolutionized by the Ponseti method with excellent results and such meddling is unwarranted.<sup>21</sup> The patients would have been better treated if they were brought to the right place.

In the present study, majority of the traditional bone setters utilized massage with herbal concoctions, splinting and bandaging although some also made scarification at site. This technique of traditional splinting of fractures is what gives rise to the gangrene. Traditional bone setters may make scarification at the site of the injury and massage the area with herbal concoctions. They then go ahead to apply splints which can be made from sticks or bamboos arranged over the site. Sometimes these sticks/bamboos are tied together. These sticks or bamboos are then tightly bound with bandage to prevent movement at the fracture site. This creates a tourniquet effect leading to vascular compromise, syndrome, compartment ischaemia and limb gangrene. 11,22-25 In addition, the scarification made predispose to infection. 11,22 Children are particularly vulnerable to the effect of this splinting as they are forced to comply with the tight splint applied by the traditional bone setter even when they feel very uncomfortable with the splint, until the limb becomes gangrenous.<sup>26</sup>

Most of the patients in the present study presented with sepsis. Sepsis often complicates the gangrene and could be a contributory factor to mortality especially if the patients present late. 9,11,25 The only mortality in the present study had septicaemia as well as generalized tetanus. Tetanus complicating lower extremity fractures and limb gangrene managed by traditional bone setters had been reported by others. 25,27

In the present study, all the patients had provisional amputation and at a later time stump refashioning with one having split skin graft. The provisional amputation removes the source of sepsis quickly in wet gangrene and reduces the inflammatory response.<sup>12</sup> However, if the gangrene is dry, it can be allowed to demarcate before the provisional amputation. <sup>1,2,5,6</sup>

The most frequent level of amputation in the present series was below knee. Although not clearly stated as a distinct entity, inferring from the level of the conditions that resulted in the lower extremity bone setters' gangrene, in the study by Akinyoola et al there were more below knee amputations also.<sup>25</sup>

In the present study, the marital status of the parents of the patients did not correlate with the amputation level. Although there were more who were married, those who were divorced/separated made up a significant percentage (28.8%). A case series by Adesina et al examined the family structure and occurrence of bone setter's gangrene in children.<sup>28</sup> They showed that children that are raised in single-mother families, step-parent families or families with discordant marriages appear prone, both to traumatic injuries and being taken to the traditional bone setter for care and ultimately developing bone setter's gangrene. This is particularly important in Nigeria with the increase in single parenthood occasioned by high rates of divorce, separation, birth to unmarried couples, desertion, economic instability and social movement that are disrupting the traditional family systems.<sup>28</sup>

In Nigeria, up to 85% of patients with fractures present first to the traditional bone setters before coming to the hospital.<sup>17</sup> Traditional bone setters' gangrene is a major contributor to indications for amputations in developing countries and especially in children.<sup>9,22-26,28</sup> The resulting loss of limb is unwarranted and is both an avoidable and preventable disaster.<sup>9,12,22-25,29</sup> Limb loss in the lower extremity can result in significant morbidity, disability and profound economic, social and psychological effects on the patient and family especially in developing countries where prosthetic services are poor.<sup>30-32</sup> This is particularly significant for children who would have to cope with these challenges for the rest of their lives.

Hence, there is a need to prevent these avoidable limb losses from traditional bone setter's gangrene. Preventive measures that have been advocated include public health education and enlightenment, education and training of traditional bone setters, prohibition of advertisement by traditional bone setters in print and electronic media, regulation of the practice of traditional bone setters, government subsidy of hospital treatment costs, expansion of the National Health Insurance Scheme to cover fracture treatment procedures and strengthening of the traditional family system. 12,13,17,18,22,24,25,28

A limitation of this study is the small number of patients. This will limit the statistical inferences that can be drawn.

#### CONCLUSION

Children at any age can develop lower extremity gangrene from the care of traditional bone setters. There was a female preponderance. The lower extremity gangrene were mainly associated with tibiofibular fractures and congenital clubfoot. A two-stage process involving initial provisional amputation and stump refashioning at a later time was adopted with good outcome. Traditional bone setter's gangrene of the lower extremity is an avoidable and preventable sequelae of traditional bone setting. There is a need to prevent this through parental health education amongst others.

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