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# Kwashi Shakes in Port Harcourt, Nigeria - A Case Report

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#### ARTICLE INFO ABSTRACT

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**Keywords:** Jerking, Kwashi Shakes, Nutritional Rehabilitation, Severe Acute Malnutrition, Trembling. **Background:** Kwashi shakes is a rare neurological complication of unknown aetiology in children with severe acute malnutrition. Previous reports show that it usually presents as coarse tremors one to eight weeks after commencement of nutritional rehabilitation in children with kwashiokor. We present a case of kwashi shakes with no prior nutritional rehabilitation. This is the first case of kwashi shakes managed in our facility.

**Methodology:** A two year old male presented with a three day history of progressive body swelling and jerky body movements. He stopped breastfeeding at one year and was fed 2-3 times daily on mainly carbohydrates and beans. On examination, he was conscious, apathetic, wasted, moderately pale with sparse fluffy brown hair and pitting pedal oedema. His weight and length were 7kg and 69cm respectively (Z-score of <-3SD for age and sex). He had continuous coarse trembling of the upper limbs and head nodding. The trembling was worse during voluntary movements and reduced and stopped during rest and sleep. His tone was reduced in all limbs. Investigations showed a packed cell volume of 26%, elevated white cell counts, positive smear for *plasmodium falciparum* and normal findings of serum glucose, calcium, electrolytes, and cerebrospinal fluid analysis. He responded to nutritional rehabilitation, antibiotics, antimalarial and multivitamins. His symptoms subsided over two weeks.

**Conclusion:** The case report shows an unusual presentation of kwashi shakes where the patient presented without any prior nutritional rehabilitation. Health care providers should be on the lookout and should not confuse it with meningoencephalitis or seizure disorder.

#### INTRODUCTION

Malnutrition still remains a problem globally but especially in developing countries like Nigeria<sup>1,2</sup>. Globally, nearly 20 million children suffer severe acute malnutrition with majority of them in Asia and Sub-Saharan Africa according to UNICEF<sup>1,2</sup>. Nearly half of all deaths in children under five years, are attributable to undernutrition. Undernutrition puts children at greater risk of dying from common infections, increases the frequency and severity of such infections, and delays recovery.<sup>1</sup> In Nigeria, 37 percent of children, or 6 million children, are stunted (chronically malnourished or low height-for-age); more than half of these severely.<sup>3</sup>

Severe acute malnutrition (SAM) is defined by a very low weight-for-height below -3

Z scores of the median World Health Organization (WHO) growth standards, manifest by visible severe wasting, or by the presence of nutritional oedema.<sup>2</sup> In children aged 6–59 months, an arm circumference less than 110 mm is also indicative of severe acute malnutrition.

The management of SAM involves initial treatment /stabilization, rehabilitation and follow-up on nutritional rehabilitation. The initial treatment/stabilization phase is to manage the immediate health problems like dehydration (fluid resuscitation using rehydration solution malnutrition - RESOMAL), infections (antibiotics, antimalarials), hypoglycaemia and initiate feeding (glucose and food such as ready-to-use-therapeuticfeeds - RUTF), temperature regulation (kangaroo mother care - KMC , extra clothing for warmth), electrolyte imbalance ( multivitamins). It is in the rehabilitation phase that iron therapy is added, and finally follow-up is continued with emotional stimulation, follow up visits, ensuring adequate weight gain. While refeeding the child, protein is added using milk, animal protein etc. Sometimes, there is aggressive protein ingestion either at the hospital or at home and these children may then rarely present with coarse jerking which imitates Parkinson's disease, in that it is worse during activity but reduces and even stops during rest and sleep. This coarse shaking was called Kwashi shakes in 1954 by Kahn in South Africa.

Kwashi shakes is a rare neurological complication of oedematous SAM that occurs during nutritional rehabilitation especially following aggressive feeding with protein especially milk<sup>4</sup>. It usually manifests as coarse jerking of the limbs, head and sometimes the abdomen and has a good prognosis resolving within a few days to weeks.<sup>4,5</sup> Kwashi shakes is also known as Parkinsonian tremors as it worsens with activities and subsides with rest. It is not known why kwashi shakes occurs in some children and not in otheres.<sup>4</sup> Kwashi shakes is usually self limiting.

It has been reported in some countries like Jamaica, South Africa and Nigeria (during the Nigerian-Biafran Civil war and Sokoto). During the Nigerian-Biafran civil war, malnourished children with

kwashiorkor were noticed to have rhythmic twitching shaking tremors, jerky movement of limbs, tongue and head 1-2 weeks following introduction of better diet.<sup>5</sup> There was no loss of consciousness and children could use the limb to play.<sup>5</sup> These children recovered within a few weeks of onset, with no neurologic sequelae<sup>5</sup>.

The jerking was different from meningitis, encephalitis and epilepsy, and all investigations including cerebrospinal fluid (CSF) analysis and electroencephalogram (EEG) were normal.<sup>4,5</sup> In Sokoto, four cases were reported and all recovered fully.<sup>5</sup>

This is the first reported case in Rivers State. In addition, this case is different in that it is occurring without a prior nutritional rehabilitation, hence a call to actively look out for it in children with SAM. We report this case to draw the attention of physicians and other health care providers to this mode of presentation.

#### **CASE PRESENTATION**

A 2 year old male presented with complains of progressive weight loss of three months duration, generalized body swelling and tremor on the upper limbs all of three days duration. Weight loss was progressive as evidenced by looseness of previously fitted clothes and child appearing smaller than peers. Body swelling was noticed by father three days prior to presentation when he went to pick up the child from the relative he had been staying with for about two months. Swelling started from the face, then the abdomen, scrotum and then legs. No history of reduction in urinary output.

Tremor was first noticed on the left hand three days prior to presentation. It was present only when patient is awake and disappeared with sleep. Tremors gradually progressed to involve the right upper limb and head. It was aggravated by voluntary movements (like feeding, lifting the hands) and reduced or disappeared with rest and sleep. At onset of symptoms, he was given multivitamins and with persistence, presented to our facility for expert management. There was no previous history of hospital admission, surgery or blood transfusion. Pregnancy was supervised in a primary health center, uneventful and neonatal period essentially normal. He was exclusively breastfed for 6 months, commenced on complementary feeds and stopped breastfeeding at one year. He eats family diet that usually consists of locally made cassava flakes (garri) and soup, rice or beans and cereals 2-3 times in a day. He took milk and eggs sparingly. He fed himself but was sometimes assisted by elder siblings and father. He was fully immunized in infancy according to National Programme on Immunization schedule and walked at one year. Patient is the youngest of 4 children whose parents separated two months prior to presentation. Father is a 30 year old mechanic apprentice with primary level of education and mother is a housewife with primary level of education. Mother abandoned the family two months prior to presentation and the children were sent to live with paternal aunt. The father went to pick them a week prior to presentation. Siblings are 11yr old female, 9yr and 6yr old males. Patient lives with the father and siblings in a one bedroom apartment and did not sleep under insecticide treated net. His source of drinking water was boiled borehole water and refuse was disposed of a few meters away from the house.

At presentation, he was moderately pale, had sparse brown fluffy hair, wasted with loss of subcutaneous fat and had bilateral pitting leg edema up to the knees. His weight and length were <-3SD on the WHO z- score (7kg and 69cm respectively). His mid upper arm circumference (MUAC) was also low at 10.5cm. He was conscious and apathetic and had continuous trembling of both hands (left more than right) with nodding of the head. Tremors were worse when he attempted voluntary movements like feeding, raising the hands or reaching out to take an object and reduced or disappeared during rest or sleep respectively. Sometimes the tremors were so severe that he had to use one limb to support the other in an

attempt to control them. His tone was reduced in all limbs and deep tendon reflexes were not exaggerated. Respiratory rate was normal at 30cpm and breath sounds were vesicular. His abdomen was distended but no organs were palpable. His heart rate was 107bpm and heart sounds 1 and 2 only were heard.

Investigations done showed a packed cell volume of 26% with elevated white blood cell count, normal blood sugar, normal serum electrolytes, urea and creatinine levels; normal calcium, potassium and magnesium levels. The cerebrospinal fluid biochemistry was normal and culture yielded no growth. Chest radiography was normal. Human immunodeficiency virus, Hepatitis B and C serology tests were non-reactive. MP showed 1+ of trophozoites of *Plasmodium falciparum*.

He was commenced on nutritional rehabilitation, antibiotics, anti-malarial drugs and responded well to treatment with gradual resolution of symptoms within two weeks.



Fig 1. Supporting the left upper limb with the right due to the trembling



Figure 2: Still able to feed himself despite the tremors. Note pedal oedema and wasting of the limbs

# **DISCUSSION**

Kwashi shakes is a rare neurological complication that occurs in children with oedematous forms of severe acute malnutrition and occurs within days to weeks of refeeding with protein-rich diet especially aggressive refeeding with milk. This child presented ab initio with kwashi shakes meaning it occurred before refeeding. His investigation results were essentially within normal limits. He responded well to treatment as expected, with no obvious neurologic sequelae.

The case presented is a two year old child. The other children affected fell between 12-24 months. The reason may be that these children are now fully introduced to family diet whose taste they are yet to getting used to. At this age also, they are more prone to infections, are unable to feed themselves properly and where there is food insufficiency they receive the least amount hence their predisposition to malnutrition. The child in our report is a male similar to the report in Jamaica as well in

Sokoto, Nigeria (3:1) in keeping with a male predilection to malnutrition in children under the age of five years. Males may therefore have a higher predilection to kwashi shakes based on being numerically more undernourished.

There is no known cause for kwashi shakes in this report although previous studies related it to a complication of nutritional rehabilitation where there was aggressive protein intake either in the hospital or at home. Although the exact cause of Kwashi shakes is not known the increase in food to a child who has been starved for sometime may be comparable to the gene thrift theory<sup>9</sup> where children who had inutero malnutrition now meet abundance postnatally and this leads to obesity and diabetes mellitus in those who are genetically predisposed. Is it possible that there may be a genetic predisposition to KS in children with In Sokoto,<sup>5</sup> one of the cases was fed 5 eggs a day by the grandmother in a bid to hasten weight gain while other mothers who had unlimited access to Ready-tofeed-therapeutic-feeds. In our case report however,

this child was yet to commence nutritional rehabilitation.

Kwashi shakes usually manifests as coarse jerking of the limbs, head and sometimes the abdomen (resembling Parkinson's disease)4-7 and this was also observed in the index patient. The patient usually assumes a characteristic posture with abduction of the shoulder joint and flexion of the elbow, and metacarpo-phalangeal joints which was seen occasionally in our patient.4 During the Nigerian-Biafran civil war, malnourished children with kwashiokor were noticed to have rhythmic twitching shaking tremors, jerky movement of limbs, tongue, head one to two weeks following introduction of better diet. It is not associated with loss of consciousness and children could even use the limb to play. The shaking is worse during activity but reduces and even stops during rest and sleep. The index patient had coarse tremors that were worsened when he tried to grab objects and disappeared when he slept.

The clinical presentation is a differential of meningitis/encephalitis hence CSF studies were done. The absence of fever in this child did not support meningitis /encephalitis as seen in other studies. There were no abnormalities in CSF findings in previous studies. The same was observed in our patient. All the patients including ours, were conscious throughout.

Other investigations to rule out other diagnosis like epilepsy include EEG which is usually normal. Brain MRI is usually normal. These investigations were not done for the index patient.

It has been noted that when serum calcium is normal there is quick resolution of kwashi shakes<sup>5</sup> just as happened in our patient. Hypocalcemic patients usually take a longer time to recover. Although the reason for delayed recovery in hypocalcemic kwashi shakes is not known we know that calcium is required for normal muscle functioning and activity by stabilizing the resting membrane potential of neurons preventing spontaneous thereby activation. Hypocalcaemia causes increased neuromusccular excitability leading to seizures, tetany, muscle fasciculations, hyperthermia stiff gait, weakness and generalized tremors 10. It is therefore possible that the presence of hypocalcaemia delayed the recovery of kwashi shakes in some patients<sup>5</sup> while in its absence recovery was faster<sup>5</sup> as in our case report.

Management includes converting the excessive protein intake<sup>4-7</sup> and ensuring the child receives only the recommended protein intake in nutritional rehabilitation and this was done for our patient.

Our patient's symptoms resolved with two weeks similar to the reports from other studies.<sup>4,6</sup> These children recovered within a few weeks of onset and no neurologic sequelae<sup>4-7</sup> just like in this patient.

#### CONCLUSION

The case report shows an unusual presentation of kwashi shakes where the patient presented without any prior nutritional rehabilitation. Health care providers should be on the lookout and should not confuse it with meningitis, encephalitis and seizure disorder.

#### What is already known on this topic?

Children with oedematous severe acute malnutrition may have neurological complications such as coarse tremors (Kwashi shakes) during recovery due to high protein diet.

#### What this study adds

Kwashi shakes may occur even before commencement of nutritional rehabilitation.

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#### Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the father of the patient has given his consent for his son's r images and other clinical information to be reported in the journal. The patient's father understands that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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#### Conflicts of interest

There are no conflicts of interest

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