



RESEARCH ARTICLE

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ENURESIS AMONG CHILDREN WITH SICKLE CELL ANAEMIA IN PORT HARCOURT

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ABSTRACT

Background: Sickle-cell anaemia (SCA) is the most common inherited haemoglobinopathy affecting the black race and enuresis have been reported to be commoner among children with SCA than those with normal haemoglobin genotype. **Aims:** The study aims to determine the prevalence of enuresis, its type, risk factors and management methods among children with SCA. **Method:** This was a cross-sectional study carried out among children with SCA on follow up at the Paediatric Haematology clinic of the University of Port Harcourt Teaching Hospital, Port Harcourt, Nigeria. A structured investigator administered questionnaire was used to obtain information on biodata and enuresis data of these patients. Obtained data were analysed and results presented in simple proportions and comparisons of subgroups carried out using the chi square test or Fischer's exact test. Statistical significance was set at p value <0.05. **Results:** A total of 96 subjects with SCA participated in the study, 52 (54.2%) were males while 44 (45.8%) were females. Thirty nine out of the 96 subjects had enuresis, giving a prevalence rate of 40.6 %. All surveyed subjects had primary nocturnal enuresis. There was a statistically significant association between enuresis and a family history of enuresis (p <0.05). Enuresis was commoner in males and decreased with increasing age, however these were not statistically significant (p >0.05). None of the subjects visited the hospital for medical treatment of enuresis. Waking child up at night, withholding water, punishment and use of herbal drugs were the common methods of enuresis management. **Conclusion:** Prevalence of nocturnal enuresis among children with SCA is high, primary nocturnal enuresis was the only type of enuresis and there is a significant association between enuresis and a positive family history of enuresis. The incorrect methods of management of enuresis requires public health enlightenment.

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INTRODUCTION

Enuresis is defined as the repeated passage of urine into clothes or beds by night or day at a voiding frequency of twice weekly for at least 3 consecutive months or the presence of significant distress or impairment in social, academic (occupational) or other important areas of functioning by a child five years or older and the behaviour not being due exclusively to the direct physiological effect of a substance (a diuretic), or a general medical condition (e.g., diabetes, spina bifida or seizure disorder), (American Psychiatric Association, 2000; Ozkan, 2007; De Sousa, 2004 and Neil, 2000). Day-time bedwetting is referred to as diurnal enuresis (DE) while nighttime bedwetting is referred to as nocturnal enuresis (NE). Combined day and night-time bedwetting is referred to as nocturnal/diurnal enuresis. NE may be primary or secondary (Robson, 2019).

Sickle-cell anaemia (SCA) is the most common inherited haemoglobinopathy which affects millions of people in the Tropics especially sub-Saharan Africa (Robert, 2007), and is also common among African-Americans (Duane, 2005). Nigeria accounts for the largest number of cases worldwide with a prevalence of about 20 per 1000 births (WHO, 2005). Children with SCA have reversibly and irreversibly sickled red blood cells, which become rigid having lost their deformability and consequently block the microvasculature resulting in vasoocclusion, ischaemia and infarction. Infarction-related renal complications start from childhood (Platt, 1994), and include tubulopathies like impaired ability to concentrate urine which manifest as hyposthenuria, polyuria and enuresis and abnormalities of urine acidification (Embury, 1995; Itsa-Rayoungyen, 1982 and De Fonzo, 1979). Studies that evaluated the prevalence of enuresis among children and adolescents with SCA show that they may be at a higher risk

of nocturnal enuresis than their counterparts with normal haemoglobin genotype (Mabiala Babela, 2004; Barakat, 2001; Jordan, 2005; Akinyanju, 1989; Ogunrinde, 2007; Ali, 2014 and Ugwu, 2007). Akinyanju *et al.* (Akinyanju, 1989) reported a high prevalence of 41.6% in South-west Nigeria among Paediatric SCA patients compared to 18.5 % in their counterparts with normal haemoglobin. A prevalence of 50.9% was reported among children with SCA in Congo Brazzaville, compared to 16.4 % among their peers with normal haemoglobin genotype, also Chizoma *et al.* (Eneh, 2015) in South East Nigeria reported a prevalence of 31.4 and 21.4 % among SCA and peers with normal haemoglobin genotype respectively, while Paul *et al.* (Paul, 2012) reported a prevalence of 23.2% among school aged children in Port Harcourt. Enuresis causes significant psychosocial problem for both parents and children especially when it persists in older children. It places a child at risk of being a target for name-calling and teasing from peers, behaviour that can damage a child's self esteem and place him or her at risk of rejection. (Robson, 2019; Paul, 2013; Gumus, 1999 and Marshall 1973). This is worse in the presence of an underlying chronic disease like SCA. The presence of enuresis can place a limit on participation in highly desirable social experiences such as holiday camps and sleepovers. These children are commonly punished and are at significant risk of emotional and physical abuse (Marshall, 1973). Studies report feeling of embarrassment, inferiority and anxiety; loss of self-esteem; and effects in self perception, interpersonal relationship and school performance (Gumus, 1999; Marshall, 1973; Kalo, 1996; Moffatt, 1989). However, there appears to be dearth of literature on the prevalence of enuresis among children with SCA in South-South Nigeria, the present study was therefore conducted to determine the prevalence of and the risk factors for nocturnal enuresis among children aged 6-16 years with SCA seen in a tertiary health facility in Port Harcourt, South-South Nigeria. It also aims at identifying common methods of management of enuresis used in these children.

MATERIALS AND METHODOLOGY

This was a cross-sectional study carried out at the weekly Paediatric Haematology clinic of the University of Port Harcourt Teaching Hospital (UPTH) over 3 months. A structured investigator administered questionnaire was used to obtain information from stable state sickle cell anaemia (SCA) patients with confirmed HbSS genotype aged 6 -16 years who were on follow up. Information obtained include the sociodemographic characteristics of the subjects, occupation and highest educational level of the parents, presence, type of, risk factors to and method of management of enuresis among these subjects. Subjects whose parents did not consent, who refused to assent to the study and those with serious medical conditions like diabetes, seizure disorder, spina bifida or who were on diuretics were excluded from the study. Enuresis in this study was defined using the DSM IV-TR criteria (Neil, 2000); which defines enuresis as the repeated passage of urine into clothes or beds by night or day at a voiding frequency of twice weekly for at least 3 consecutive months by a child five years or older and not being due to a substance (e.g. a diuretic), or a general medical condition (e.g., diabetes, spin a bifida or seizure disorder).. Social class of the subjects was determined based on the socio-economic class described by Oyedeji; (Oyedeji, 1985) which uses the occupation and educational level of the parents. The occupation and the educational attainment of the parents were used to determine

the socio-economic index scores of the subjects. Each subject was assigned scores based on parent's (father's and mother's) education and occupation. The scores (two for the father and two for the mother) were summed up and the mean (approximated to the nearest whole number) obtained. The mean of four scores was used to assign the subject to one of the socioeconomic groups (I-V). Obtained data were entered in Microsoft Excel and descriptive statistical analysis was performed and results presented in prose and as frequency tables simple proportions and comparisons of subgroups carried out using the chi square test or Fischer's exact test. Statistical significance was declared at p value <0.05. Ethical approval was from the hospital Research and Ethics Committee as well as verbal consent and assent from the caregivers and the subjects.

RESULTS

Age and Sex distribution of the study population: A total of 96 subjects with SCA participated in the study. Of this, 52 (54.2) were males while 44 (45.8%) were females. There was a good distribution of the study participants between the sexes was not statistically significant Table 1.

Table 1. Age and Sex distribution of the study population

Age group(yrs)	Males (%)	Females (%)	Total (%)	Chi-square (p-value)
6 -8	15 (28.8)	12(27.3)	27(28.1)	0.59 (0.9634)**
9-11	14 (26.9)	13 (29.5)	27 (28.1)	
12 -14	12 (23.1)	12(27.3)	24 (25.0)	
>14	11 (21.2)	7 (15.9)	18 (18.2)	
Total	52 (54.2)	44 (45.8)	96 (100.0)	

Prevalence/type of Enuresis: Out of the 96 subjects, 39 of them had enuresis, giving a prevalence rate of 40.6 %. More males (61.5%) than females (38.5%) were found to be enuretic (male: female ratio of 1.6: 1). The observed sex difference was not statistically significant (p=0.5161), (Table 2). All surveyed subjects had primary nocturnal enuresis.

Table 2. Prevalence of Enuresis

Variable	Males (%)	Females (%)	Total (%)	Chi-square (p-value)
A	24 (61.5)	15 (38.5)	39 (40.6)	0.42
B	28 (49.1)	29 (50.9)	57 (59.4)	(0.5161)**
Total	52 (100.0)	44 (100.0)	96 (100.0)	

Prevalence of enuresis by age: The prevalence of enuresis decreased with increasing age. Eleven (28.2%) of the children aged 6-8 years were enuretic, while 9 (23.1%) of those aged more than 14 years were enuretic. However, there was no statistically significant relationship between the age of the patients and enuresis (p=0.8261) Table 3.

Table 3. Prevalence of enuresis by age

Age (years)	Enuresis			Chi-square (p-value)
	Yes (%)	No (%)	Total (%)	
6-8	11 (28.2)	16 (28.1)	27 (28.1)	0.89 (0.8261)
9-11	10 (25.6)	17 (29.8)	27 (28.1)	
12-14	9(23.1)	15 (26.3)	24 (25.0)	
>14	9 (23.1)	9 (15.8)	18 (18.8)	
Total	39(40.6)	57 (59.4)	96 (100.0)	

Age distribution of prevalence of enuresis by Gender: The prevalence of enuresis decreased with increasing age in both sexes as shown in Table 4.

studies and exceeds population prevalence and that found in children with homozygous HbAA genotype. Ogunrinde *et al* (Ogunrinde, 2007), reported a prevalence of 40.7% among

Table 4. Age distribution of prevalence of enuresis by Gender

Age (Years)	Enuresis (Males)			Enuresis (Females)		
	Yes (%)	No (%)	Total (%)	Yes (%)	No (%)	Total (%)
6-8	7 (29.2)	8 (28.6)	15 (28.8)	4(26.7)	8 (27.6)	12 (27.3)
9-11	6 (25.0)	8 (28.6)	14 (26.9)	4 (26.7)	7 (24.1)	13 (29.5)
12-14	5 (20.8)	7 (25.0)	12 (23.1)	4 (26.6)	8 (27.5)	12 (27.3)
>14	6 (25.0)	5 (17.8)	11 (21.2)	3 (20.0)	4 (13.8)	7 (15.9)
Total	24(46.1)	28 (53.8)	52 (100.0)	15 (34.1)	29 (65.9)	44 (100.0)

Table 5. Enuresis and social class

Social class	Enuresis			Chi-square (p-value)
	Yes (%)	No (%)	Total	
I-II (upper)	10 (25.6)	14 (24.6)	24 (25.0)	0.01 (0.9914)**
III (Middle)	13 (33.3)	19 (33.3)	30 (31.3)	
IV-V (Lower)	16 (41.0)	24 (42.1)	42 (43.7)	
Total	39 (40.6)	57 (59.4)	96(100.0)	

Table 6. Enuresis and positive family history

Positive family history	Enuresis		Total (%)	Fischer's Exact
	Yes (%)	No (%)		
Yes	25 (64.1)	8 (14.0)	33 (34.4)	0.0001
No	11 (28.2)	49 (86.0)	60 (62.5)	
No response	3 (7.7)	0 (0.0)	3 (3.1)	
Total	39 (100.0)	57 (100)	96 (100.0)	

Table 7. Methods of management of enuresis in the study group.

Treatment received	No	Percentage (%)
Waking up at night	37	94.9
No drinking of water in the night	14	35.9
Punishment	9	23.1
Traditional Herbs	8	20.5
Nothing (waiting for Child to outgrow it)	5	12.8
Medical treatment	0	0.0

Enuresis and social class: Of the 96 subjects who were enuretic, 16 (41.0%) belonged to the lower social class while 13 (33.3%) were from the middle social class. There was no statistically significant association between enuresis and social class as shown in Table V.

Enuresis and positive family history: Among those who were enuretic, 25 (64.1%) had a positive family history of enuresis, while 8 (14.0%) had a similar history among those who were not enuretic. The observed difference was found to be statistically significant ($P=0.00$). There was a positive family history of enuresis in 28 (71.8%) of fathers, in 7 (17.9%) of mothers and in 4(10.3%) of siblings among those who were enuretic. There was a positive history of enuresis in both the mother and father in 21 (53.8%) of those who were enuretic.

Methods of management of enuresis in the study group: Table 7 shows that none of the subjects presented to the hospital for medical treatment. Thirty seven (94.9%) of the subjects were awakened at different points in the night to urinate while 9(23.1%) were punished for bed wetting.

DISCUSSION

The prevalence of nocturnal enuresis among patients with SCA found in this study compares to those reported in other similar

patients with SCA while Akinyanju *et al.* (1989) and Ali *et al.* (2014) reported similar prevalence of 41.2 and 48.3% among children with SCA respectively. These authors and others however, reported lower prevalence ranging from 17.7 -23.2% (Akinyanju, 1989; Ogunrinde, 2007; Ali, 2014; Paul, 2012). among non SCA patients and in population studies. The definitive cause of increased nocturnal enuresis among patients with SCA is not known; increased urinary volume resulting from a low urine concentrating ability of the kidneys and hyposthenuria that occurs early in childhood in many of these children may not be the sole factor as it cannot explain why some children are spared. It is also possible that the degree of renal affectation in these patients vary in severity and hence the disparity. Also, maturational delay has been implicated in the aetiology of nocturnal enuresis and this may play a role among SCA patients. Delay in pubertal changes is a known fact among these children and this may be related to SCA related brain injury where the micturition control centre is located (Adams, 2020 and Blok, 1994). The risk factor to enuresis among children with SCA is probably multifactorial. In this study, more males than females were found to be enuretic though this was not statistically significant. Male preponderance of nocturnal enuresis in children with SCA have been reported from other studies (Barakat, 2001; Akinyanju, 1989; Ogunrinde, 2007; Anochie, 2006; Ozden, 2007; Readett, 1990).

A similar trend has also been reported among children with normal haemoglobin genotype (Ogunrinde, 2007; Chiozza, 1988; Golding, 1986; Garfinkel, 2000 and Paul, 2013). This suggests that factors contributing to enuresis in normal children such as slower maturation and reduced responsiveness to toilet training in boys (Golding, 1986), as well as more frequent developmental delays (Garfinkel, 2000), are equally important in children with SCA. In contrast, other researchers have documented a female predominance in their subjects (Mabiala Babela, 2004). Generally, limited progress has been made in establishing the aetiology and pathophysiology of nocturnal enuresis in subjects with SCA. The prevalence of enuresis in this study decreased with increasing age and this was true with both sexes, however it was not statistically significant. Decreased prevalence of enuresis with increasing age and vice versa is a common phenomenon reported among SCA patients and in the general population. This is also probably related to the maturation delay theory in its aetiology as there is a spontaneous resolution rate of 15% per year (that is, with increasing age some enuretics stop bedwetting without treatment) with increasing age (Robson, 2019; Ogunrinde, 2007; Paul, 2013).

In the present study, it was noted that socio-economic status did not have any significant association on the prevalence of nocturnal enuresis and this is in tandem with the findings by other authors within and outside Nigeria (Ogunrinde, 2007; Anochie, 2006; Readett, 1990; Paul, 2013). This finding is however at variance with previous observations (Chiozza, 1988; Golding, 1986; Garfinkel, 2000), which noted that enuresis was more frequent among normal children from the lower socio-economic class. Conversely, a study in Benin city, Nigeria reported nocturnal enuresis to be commoner among normal children from higher socio-economic class (Obi, 1977). Therefore, the role of socioeconomic status as a risk factor to nocturnal enuresis is uncertain. Readett and co-workers attributed this finding to the relative unreliability of social amenities in predicting social status (Readett, 1990). Many studies (American Psychiatric Association, 2000; Ogunrinde, 2007; Eneh, 2015; Paul, 2013; Azhir, 2006) have reported an underlying genetic risk to enuresis in both SCA and the general population, but this genetic factor is yet to be identified as different foci has been linked to it. It is possible that the factor that operates at the general population also plays a role among children with SCA. In this study, in keeping with the proposed genetic basis of enuresis, many enuretic children (64.1%) had a positive family history of enuresis compared to 14.0% in the non enuretic children and this difference was statistically significant relationship A positive history in the fathers seem to have a dominant role compared to that of mothers and siblings and this has been reported in some studies (Azhir, 2006).

Treatment of nocturnal enuresis minimizes the embarrassment and anxiety of the child and the frustration experienced by the parents, it also improves and preserves the child's self-esteem (Robson, 2019). Studies have shown restoration and new sense of confidence, improvement in self-esteem and academic performance, positive personality change and improvement in family relationship in treated enuretics. In this study, none of the enuretics received medical treatment; this is because many parents believe that enuresis is always self-limiting and that nothing can be done medically and they must wait for their child to grow out of it, while others fail to report enuresis due to the embarrassment it may cause for their child. As in most

previous studies, the rates of consultation for the management of enuresis were low all over the world ranging from 0-38%. (Robson, 2019; Ogunrinde, 2007; Golding, 1986; Paul, 2013). In the current study, the children were punished in 23.1% of the cases, many of the parents practiced waking the enuretics up at night, depriving them of water in the nights and use of herbal medications. These methods have not been shown to be effective and may lead to dehydration among SCA whose urine production is often high. Studies have shown that punishment is not entertained in the management of enuresis because they adversely affect the psychological development of the child and may worsen it in those with an already existing negative psychology (Akinyanju, 1989; Ogunrinde, 2007). In conclusion, the prevalence of enuresis among children with SCA is high, Primary nocturnal enuresis was the only type of enuresis found in this study and there is a significant association between enuresis and a positive family history of enuresis. The wrong methods of management of enuresis requires public health enlightenment.

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