Niger J Paed 2015; 42 (1): 55 - 58

ORIGINAL

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The social-economic and family background of the child with a CNS birth defect in a developing country in the current era

DOI:http://dx.doi.org/10.4314/njp.v42i1,12

Accepted: 4th November 2014

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Abstract: *Objectives:* In much older literature many socio-cultural factors militating against the optimal clinical / surgical care of CNS birth defects in the low-middle income countries (LMICs) were reported. We set out to interrogate this phenomenon in the current era
Methods: A retrospective cross-sectional survey of a prospective data-base of the social-economic and family background of the children with CNS birth defects presenting for surgical care in a busy neurosurgical practice in Nigeria.
Results: There were 151 children, 81 males (53.6%), with hydrocephalus and neural tube closure defects (NTDs) seen in the study period; median age at presentation was at 4 weeks of life, the NTDs presenting much earlier than hydrocephalus, p-value< 0.001; each child represented the first of the parents in about a third of cases, and at least the 3rd or higher birth order in 40.4%. The parents were young adults, but the mean age of the fathers, 35.8years, was higher than the mothers', 30.0years, p-value<0.001; the parents had low level of education and socio-economic statuses; more than 90% had no knowledge about any preventive measures for CNS birth defects; and, in spite of their already sizeable families each, 56% of mothers, and 62% of the fathers were still gearing up for further pregnancies.
Conclusions: Coupled with the region's well-known harsh health system, the socio-economic and family background of the child with CNS birth defects remain very challenging indeed in this typical developing country. This calls for concerted efforts to promote in the LMICs the adoption of the established measures of preventing CNS birth defects.

Key words: CNS birth defects; social-economic, family background; low-middle income countries.

Introduction

Central nervous system (CNS) birth defects are among the few systemic congenital anomalies compatible with life. They have a high prevalence in the low-middle income countries, LMICs, otherwise known as the developing countries of the world. For instance, some 30% of neonatal deaths related to visible congenital anomalies in the LMICs are CNS birth defects[1]. But the cost of their immediate postnatal medical care and surgical treatment is very staggering indeed[2]. Even more so is the cost of their life-long care[3-5]. In the LMICs, a very sizeable proportion of the population lives below the national poverty line. This proportion is at least 46% in Nigeria[6]. Furthermore, the health system of much of the LMICs is privately funded in most places, meaning that the individuals pay out of pocket for all aspects of health care at the points of the service. The government / private financing of health care ratio, again, in Nigeria for instance is currently 31/69[7].

It is into this family socio-economic milieu that the child with the CNS birth defect arrives, and is presented for neurosurgical care in our practice.
This study was carried out to objectively characterize this family background. It is hoped that this exercise would help cast in bold relief the relevant socio-economic factors impacting the clinical / neurosurgical care of these major health needs in this setting.

Materials and Methods

This was a retrospective descriptive analysis of a prospective data base of all the cases of CNS birth defects

seen in the neurosurgeon's practice over the duration of four years: from May 2009 till June 2013. . The neurosurgical unit of the principal author's, a 4-faculty practice, is arguably the busiest in the country. The relevant data were continuously captured prospectively in clinical summary forms and an electronic spreadsheet. Information in each proforma usually consisted of the clinical presentation, the neurosurgical in-hospital course, post-admission outpatient follow-up of each child, and some socio-demographic data on each child's mother and father. Both parents were usually interviewed together except when one of them was not available. In that case, as much information as possible about the absent parent was obtained from the one present.

For this study the specific information extracted from this data-base included (i) the types of the CNS anomalies seen in the children; the age at presentation for neurosurgical care; the number of children in each patient's respective families, and the child's birth order, and (ii) the ages of the parents, and their respective socio-economic levels, including the maximum formal educational attainments. The parents were also surveyed on their prior knowledge of preventive measures for CNS birth defects. Finally their opinions were sampled about their readiness or otherwise to try to get pregnant again after the index child with the CNS birth defect

This data-base was analysed using the SPSS version 18 (SPSS, Inc, IL). Descriptive data are presented in frequencies / proportions, means (±standard deviations, SD) and medians. Tests of associations were performed for categorical variables using the Pearson's Chi-square (or Fisher's exact) test; with one-sample t-test for the continuous, normal-distribution variable regarding the mean ages of the fathers and mothers; and with the Wilcoxon Mann-Whitney U test for non-parametric variable with skewed distribution. The level of statistical significance was set at an alpha level of < 0.05.

Results

One hundred and fifty one children with CNS birth defects were evaluated in this study period, table 1. There were 81 males (53.6%) and 70 females (46.4%), male: female ratio 1.2: 1.

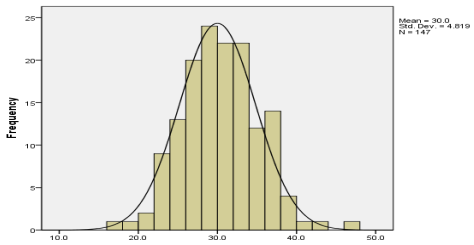
The ages at presentation for neurosurgical attention ranged from day 1 to 5110, mean 198.9 median 28.0 days; or stated in weeks, 1-728 weeks, mean 23.5, median 4.0. Further analysis showed that only 38 (25.2%) cases presented within the first week of life; or 24 (15.9%) within 72 hours of birth; and only 7 cases (4.6%) on the day of birth. About two-thirds were neural tube closure defects, NTDs; the rest were cases of hydrocephalus. The median time to neurosurgical attention of the NTDs was very significantly much shorter than the cases with hydrocephalus; $p < 0.001$ (Mann Whitney U, 1166.00; Wilcoxon W, 4736.00; $z = -6.24$). The median number of children in the families concerned was 2, range 1-11. At least one-third of this cohort represented the very first child of the respective family; 61 cases (40.4%) represented the 3rd or more child of the family.

Table 1: Types of CNS birth defects

Variables, n-151	No (%)	p-value
The CNS anomalies		
Craniospinal dysraphism	101 (66.9)	
Spina bifida	84	
Encephalocoele	17	
Hyrocephalus	50 (33.1)	
Median age at neurosurgical presentation		
Craniospinal dysraphism	14 days	
Hydrocephalus	120 days	< 0.001*
Number of Children in the family		
Range	1-11	
Median	2	
Child's birth order		
First	51 (33.8)	
Second or third	67 (44.4)	
Fourth or higher	33 (21.8)	

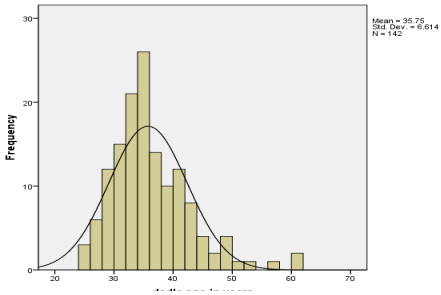
Mann-Whitney U test

Fig 1a



Mothers age in years

Fig 1b



Fathers age in years

Table 2 shows some of the relevant socio- demographic details of the parents of the study subjects. Both parents were essentially young persons in the majority, figure 1, although the fathers were significantly the older of the two: mean fathers' age 35.8 (±6.61)years, range 25-60, median 35.0; and the mothers, 30.0 (±4.82)years, range 17-46, median 30.0, $p < 0.001$. In the vast majority of them, 96.7%, the mothers either lived on low income (traders, artisans, low-earning civil servants) or had no personal source of income whatsoever: housewives, students, unemployed and so on. The fathers, though with a significantly higher proportion of high income level, p-value < 0.001, still were also of the nil / low income status in 85%. These parents also both had only basic literacy education in more than 50% each.

There was no prior family history of a child with a CNS birth defect in any of the cases; and both of the parents in the majority, >90% each, had no prior knowledge of any preventive measures for CNS birth defects. About 56% of the mothers, and 62% of the fathers, would still try to achieve further conception after taking care of the current child with the birth defect. This difference was not statistically significant.

Table 2: Socio-demographic characteristics of the parents of this cohort of children with CNS birth defects

Variable	Mothers	Fathers	p-value
Mean age of parents (years)	30.00	35.75	<0.001*
Parents' income level	Mothers n-151	Fathers n-150	
No income	29 (19.2)	8 (5.3)	
Low income	117 (77.5)	120 (80.0)	
High income	05 (3.3)	22 (14.7)	<0.001**
Parents' educational attainment	Mothers n-151	Fathers, n-142	
Primary school or less	19 (12.6)	11 (7.7)	
Secondary school	62 (41.1)	61 (43.0)	
Tertiary school	70 (46.6)	70 (49.3)	0.39**
Prior knowledge about preventive measures	Mothers n-149	Fathers n-109	
Yes	13 (8.7)	9 (8.3)	
No	136 (91.3)	100 (91.7)	0.89**
Attitude to further pregnancy	Mothers n-151	Fathers n-110	
Afraid / no more pregnancy	67 (44.4)	42 (38.2)	
Want future pregnancy	84 (55.6)	68 (61.8)	0.32**

*One-sample t-test
**Pearson's Chi Square test

Discussion

This study is a descriptive cross-sectional one briefly surveying the salient social-economic milieu, in the current era, of the typical family of the child with a CNS birth defect in a developing country. The findings of this study corroborate, even consolidate, some of the hard facts on this issue only alluded to in some previous studies[8-11]. It shows that the birth prevalence of CNS congenital anomalies is far from abating in this country; that the family milieu, ditto the health systems, to which the children with these defects are born is one that is actually ill-positioned to offer them the dedicated, capital-intensive immediate and long-term social-medical care needed[12,13]. Majority of the parents earned low income, or nil whatsoever; had only low-level educational attainment, and already had sizeable families each[14]. It would be imagined, for practical purposes, that it could only be hard struggle indeed for a poor family already with 1 or 2 children to be saddled with an additional healthy child, not to talk of one with a CNS birth defect.

Congenital anomalies of the CNS are among the few birth defects that are compatible with life. They are however more devastating than the rest of the birth defects in many ways. The cost of their immediate postnatal medical care and surgical corrections can be very staggering indeed, even for the well-endowed health systems[3]. And that is usually just the beginning of the story. The life-long social, economic, and even personal / family burdens, associated with living with them are simply unquantifiable[3,4,13].
Here, therefore, are some of the paradoxes of the fact of CNS birth defects in the LMICs. They are a disease which even the rich families cannot easily afford the care of, yet they affect in the main the struggling, impoverished members of the population. The immediate postnatal medical care, and surgical corrections, of these defects can be very complex indeed, needing well funded, cutting-edge medical practice, yet the reverse is the reality of the health systems of much of the LMICs[1]. Here, the health system as a whole is a very harsh one indeed. It is essentially unorganized, able to sustain only elementary health care at best, and is poorly funded by the government. Thus private funding of health care in these regions averages 70% to the government's 30%, a complete reversal of the private /government funding ratio of health care in the advanced countries of the world[7]. It has been observed actually that the out-of-pocket, point-of-service payment for basic health care needs drive some 250 million people yearly to extreme poverty and severe financial hardships in the LMICs[15].

Many more psychosocial problems are also known to attend the presence of a child with CNS birth defect in a family[13,15,16]. To the detriment of the other children in the family the whole parental financial and loving attention, at home and for hospital attendance, may be devoted to the affected child[12,13,17]. Parents may experience great anguish and, in some occasions, even feelings of personal guilt for the sufferings, real and imagined, of the affected child. Finally, occasional squabbles, and family breakups, can occur[18,19].
Thus the usual story of CNS birth defects in LMICs is usually that of late neurosurgical presentation, sometimes following an initial search for 'cheaper' alternative medicine care; suboptimal in-hospital medical and neurosurgical care, and an in-adequate / non-existing long-term organized rehabilitative care[10,20-22].

This study also reveals one more sobering paradoxical point. This is the fact that, in much of the LMICs, the hope of any short-term reduction in the birth prevalence of CNS birth defects may be a tall one indeed. For although the children affected with these devastating anomalies arrived in poor, peasant families who already had more than two children in more than 40% of the cases, a significant proportion of the concerned parents were still gearing up for more future conceptions. The risks of recurrence of NTDs, for instance, are about 10-30 times the general population[23]. And these defects are, to start with, diseases of the poor as a matter of fact[1,24]. In addition, more than 90% of the parents of the children in this study actually showed no evidence of any knowledge of the preventive measures for the CNS birth defects. There is therefore a great need for renewed efforts at primary prevention of CNS birth defects in the developing country. The issue of responsible family size and spacing should also be addressed.

Conclusions

Just as in the earlier, much dated literature on the subject from the region, the socio-economic and family background of the child with a CNS birth defect in Nigeria remains very challenging indeed[9,11,12,21,25]. The health

system is harsh. The parents are young and poor; already have sizeable families and, lack knowledge of the measures for preventing these devastating birth defects.

Authors' contributions
Each author made substantial contributions towards the conception, data gathering and analysis, manuscript drafts, and final approval for submission for publication please.
Conflict of interest: None
Funding: None

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