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The prevalence of noma in northwest Nigeria

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ABSTRACT

Background Noma, a rapidly progressing infection of the oral cavity, mainly affects children. The true burden is unknown. This study reports estimated noma prevalence in children in northwest Nigeria.

Methods Oral screening was performed on all ≤15 year olds, with caretaker consent, in selected households during this cross-sectional survey. Noma stages were classified using WHO criteria and caretakers answered survey questions. The prevalence of noma was estimated stratified by age group (0–5 and 6–15 years). Factors associated with noma were estimated using logistic regression.

Results A total of 177 clusters, 3499 households and 7122 children were included. In this sample, 4239 (59.8%) were 0–5 years and 3692 (52.1%) were female. Simple gingivitis was identified in 3.1% (n=181; 95% CI 2.6 to 3.8), acute necrotising gingivitis in 0.1% (n=10; CI 0.1 to 0.3) and oedema in 0.05% (n=3; CI 0.02 to 0.2). No cases of late-stage noma were detected. Multivariable analysis in the group aged 0–5 years showed having a well as the drinking water source (adjusted odds ratio (aOR) 2.1; CI 1.2 to 3.6) and being aged 3–5 years (aOR 3.9; CI 2.1 to 7.8) was associated with being a noma case. In 6–15 year olds, being male (aOR 1.5; CI 1.0 to 2.2) was associated with being a noma case and preparing pap once or more per week (aOR 0.4; CI 0.2 to 0.8) was associated with not having noma. We estimated that 129120 (CI 105294 to 152 947) individuals <15 years of age would have any stage of noma at the time of the survey within the two states. Most of these cases (93%; n=120 082) would be children with simple gingivitis.

Conclusions Our study identified a high prevalence of children at risk of developing advanced noma. This disease is important but neglected and therefore merits inclusion in the WHO neglected tropical diseases list.

INTRODUCTION

Noma, also known as cancrum oris, is a poorly understood, rapidly progressing infection of the oral cavity, with a reported 90% mortality rate.[1] If untreated, death usually occurs within 2 weeks after the onset of acute necrotising

Key questions

What is already known?
► Our understanding of the current disease burden and epidemiology is limited; the WHO estimates 770 000 people are currently living with noma globally.
► Three Nigerian studies estimated the burden of disease ranging from 7 cases per 1000 children aged between 1 and 16 years (2003) to 6.4 per 1000 children (2003) to 1.6 per 100 000 population at risk (2010–2018).

What are the new findings?
► The prevalence of any stage of noma was identified in 3.3% of sampled children.
► Having a well as a drinking water source, being aged between 3 and 5 years and preparing pap less than once a week were associated with higher noma prevalence.

What do the new findings imply?
► Noma is a disease with considerable burden in northwest Nigeria.
► Resource allocation to improve health systems to prevent, detect and treat noma is required and this could be enhanced if noma were added to WHO's list of neglected tropical diseases.

ulcerative gingivitis (stage 1 noma).[1 2] Treatment with antibiotics, wound debridement and nutritional support in the early reversible stages of the disease greatly reduce mortality and morbidity.[2] Noma mostly affects children aged 2–5 years, and those who survive have severe facial disfigurements and multiple functional impairments including difficulties eating, seeing and breathing, contributing towards stigmatisation.[2] Noma starts as an inflammation of the gums leading to the rapid destruction of the hard and soft tissues of the face usually within 1 week.[3] The WHO has classified noma into stages[1]: stage 0, simple gingivitis; stage 1, acute necrotising ulcerative

gingivitis; stage 2, oedema; stage 3, gangrene; stage 4, scarring; stage 5, sequelae. It is unknown what proportion of simple gingivitis cases progress to the later stages of noma, but it is thought to be a small fraction.[1] In the majority of cases, infection causes the destruction of the cheek, while destruction of the jaw, lip, nose and eye have also been reported.[4] Noma can become inactive with, and sometimes without, treatment. Once this occurs, patients can survive into adulthood but often require extensive reconstructive surgery and physiotherapy to correct the resulting defects and improve function.[1] The aetiology of noma is unknown but thought to be multifactorial.[2] Noma typifies the complex interactions between extreme poverty, malnutrition, poor oral hygiene, poor access to routine childhood vaccinations, limited access to quality healthcare and immunosuppression resulting from comorbidities such as HIV.[2]

In the 1800s, noma was widely reported in Europe[5] but is currently thought to be most prevalent in low-resource settings in Africa and Asia.[6] Based on expert opinion, the WHO estimates that 770000 people are currently living with noma globally; however, it is unclear what stages of noma are included in this estimate.[7] The oldest estimate of the burden of this disease that we could locate was from Edinburgh, UK, which indicated that noma was diagnosed once out of every 5000 cases of children with an illness between 1860 and 1871.[8] Two recent Nigerian studies estimated the burden of disease ranged from 7 cases per 1000 children aged between 1 and 16 years (2003)[9] to 6.4 per 1000 children (2003).[10] A study from 2019 estimated the period prevalence of noma from 2010 to 2018 was 1.6 per 100 000 population at risk in Nigeria.[11] These estimates are based on expert opinion, number of hospital admissions and retrospectively collected data, and it is unclear which stages of noma were included.[12] Our understanding of the current disease burden and epidemiology thus remain limited. There are few studies not only on the burden of disease but also on the pathogenesis and mortality rate. Although these aspects highlight the neglected nature of the disease, noma is not currently on the WHO neglected tropical diseases list.

Noma cases are frequently reported in Nigeria.[9 13–15] The Nigerian Centre for Disease Control recorded 37 646 noma cases from 2011 to 2017.[16] However, these records may underestimate the true burden of cases, given limited surveillance data and the potential for under-reporting (low rates of diagnosis, patients not accessing healthcare, reported high and rapid mortality).[16] The majority of noma cases are reported from the northwest and northeast of the country.[17] At the 2018 National Noma Day Workshop, the Nigerian Ministry of Health confirmed that noma was a national public health priority, and highlighted the urgent need to generate robust evidence on the country's disease burden for programmatic planning.[18] This study contributes towards this need by estimating the prevalence of noma in northwest Nigeria.
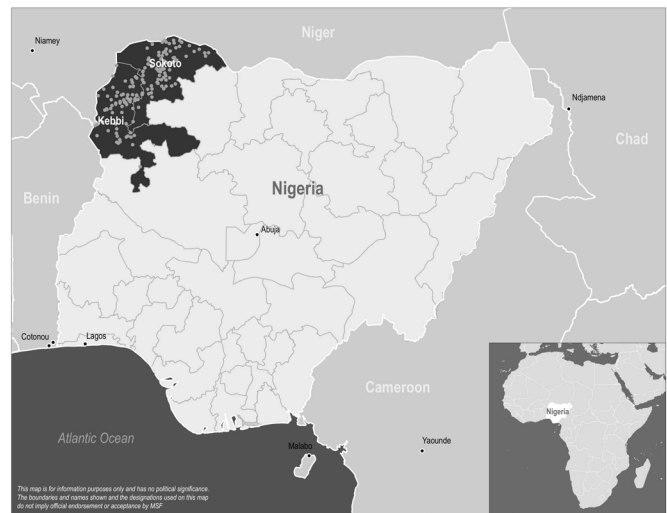
Figure 1 Map of Africa (inset) and Nigeria (main panel). Main panel, grey lines represent state boundaries. Sokoto and Kebbi states, locations for this study, are shaded dark. Pale grey dots within Sokoto and Kebbi states indicate the clusters where data collection occurred during the survey.

METHODS

Study design and setting

A two-stage cluster-based cross-sectional survey was conducted in Sokoto and Kebbi States in northwest Nigeria (figure 1).

Sampling

Sokoto and Kebbi States have estimated populations of 4 798 979 and 4 203 978, respectively.[19] Sample size calculations indicated the need for inclusion of 3615 households across 181 clusters with 20 households per cluster in order to estimate noma prevalence with precision of 0.4%. This calculation was based on the following assumptions: prior prevalence estimate, 1%[10]; design effect, 2; 1.98 children per household in the group aged 0 to 4 years[20]; average household size, 6[21]; and a 10% non-response rate.

The number of villages (clusters) per ward was selected proportional to the population size of each administrative ward. A sampling frame of villages was created by ward in Sokoto and Kebbi using geosampling remote sensing methods. The OpenStreetMap database was compared against freely available satellite imagery to identify and verify village geolocations and add new village geolocations to the list. Villages were each assigned a number and a random selection was conducted.

Study participants

All children aged ≤15 years who lived in a selected household in sampled clusters were included in the study.

Data collection

Five research teams, each with five team members, of whom one was a nurse or doctor, and one the team leader, carried out data collection. Teams were trained for 1 week prior to the commencement of data collection. Teams followed directions to selected clusters

using a mobile mapping application (OsmAnd) on data collection tablets (electronic mobile devices). Random household selection at cluster level was done using the adapted WHO Expanded Programme on Immunisation method.[22]

In selected households, consenting caretakers answered a structured questionnaire, collected on tablets using KoBoCollect (KoboToolBox), which covered sociodemographic characteristics, living conditions, child's vaccination status, oral hygiene practices, food preparation, feeding practices and access to healthcare in the 12 months preceding the interview. For the questions around feeding practices, pap was defined in this context as a type of porridge staple made from maize, sorghum or millet. Interviews were conducted in Hausa, and answers were coded automatically on the KoBoCollect tool into English.

In the sampled households, all eligible children underwent oral screening, which involved visual examination by a medical team member for any noma stage, based on the WHO classification.[1] The caretakers of children with simple gingivitis were advised to follow a strict oral health regimen (gargle with salt water or use water to clean mouth twice or more a day) as were acute necrotising gingivitis cases who were also referred to the closest health centre. If children were identified as having any later stage of noma, they were referred directly to the Noma Children's Hospital for care.

To assess the malnutrition status in children aged 6 months to 5 years, mid-upper arm circumference (MUAC) measurements were conducted using a flexible MUAC device with a precision of 1 mm.

Medical data (oral screening and MUAC) were collected on paper and later entered into a password-protected database by the study team.

Collected data were screened daily by the research team supervisors to identify inconsistencies and missing items, and immediate feedback was given to the data collection teams.

Statistical analysis

We performed descriptive analyses of household characteristics in the study sample. Categorical variables are reported as frequencies and percentages. Continuous variables are summarised using medians and IQR. Missing data numbers are recorded in each table.

Wealth scores were calculated by assigning a value of one to each of the following items owned by the family: a mobile phone, motorbike, tractor and camel (these items were chosen based on consultation with local researchers, knowledgeable about the context). The minimum wealth score was zero and the maximum was four.

Weighted prevalence and 95% CI for all WHO noma stages were estimated and stratified by age group (0–5 years and 6–15 years). The number of individuals with noma in Sokoto and Kebbi States was calculated by extrapolating the percentage prevalence from our study results to the total population in the group aged 0–15 years for

these states. This calculation took into consideration the cluster survey design and population age distribution of the two states. Using MUAC measurements, we estimated the weighted prevalence of severe acute malnutrition (SAM, MUAC <115 mm), moderate acute malnutrition (MAM, between MUAC ≥115 and <125 mm) and global acute malnutrition (GAM, MUAC <125 mm) in children aged 6 months to 5 years. The SEs of the estimates were adjusted using the linearisation method (syvset suite of Stata commands) to reflect the two-stage clustered design of the survey.[23] The estimates and SEs were weighted to account for the actual population distribution of the two states, as our survey sample was observed to have under-represented participants aged 6–15 years, when we compared our sample's age distribution with the population age distribution. The design effect (DEFF) was calculated to assess the ratio of variance under the sampling method used, in comparison to the variance of a simple random sample. This reflects the impact of the cluster sampling strategy. DEFF is reported for each prevalence and malnutrition estimate.

Univariable analysis with logistic regression was conducted to identify factors associated with noma stages 1 and 2 in the total study sample, where the number of noma cases were too small to allow for multivariable analysis.

Univariable and multivariable analyses were conducted using logistic regression to estimate factors associated with any noma stage (stage 0–2); stratified by age group (0–5 and 6–15 years). Variables chosen for inclusion in the multivariable analysis were those with 10 or more cases[24] and a univariable strength of association equivalent to a p<0.2, after assessing collinearity among variables. To further understand the association with age, an age covariate with finer age categories (0–2 years, and 3–5 years, in the younger age group model; and 6–10 years, and 11–15 years, in the older age group model, respectively) were included in the univariable analyses for both age group models, and in the 0–5 year old multivariable model.

All data analysis was conducted with Stata V.15 (StataCorp LP, College Station, Texas, USA).

Patient and public involvement

Patients and the public were involved in the framing of the study questionnaire and data collection. Dissemination of results to patients and the public will take place through outreach activities from the NCH.

Ethical considerations

Written informed consent was obtained from all literate caretakers; caretakers with insufficient literacy provided a thumbprint and a signature from a literate witness. For individuals aged 8–17 years, the child provided assent and a caretaker provided written consent.

RESULTS

The survey was conducted from 17 September to 5 November 2018, and included 3499 households in 177

clusters, 92 clusters from Sokoto and 85 from Kebbi (four clusters were not accessible because of security issues), with 7164 children aged <15 years. As 42 children did not have oral examinations, they were excluded from the analysis and the remaining 7122 were included. The median caretaker age was 30 years (IQR 25–35); 3423 caretakers (97.8%) were female; 2194 (30.8%) were employed or self-employed, and the median household size was five people (IQR 4–7). Most children (n=4239; 59.5%) were aged 0–5 years, 3692 (52.1%) were female, 5875 (83.0%) had no education, and 6686 (94.4%) had a primary caretaker that was the mother (table 1).

Prevalence

Table 2 reports the prevalence of all stages of noma in the study population overall and by age group. Any stage of noma was identified in 3.3% of sampled children (n=194; CI 2.7 to 4.0). Stage 0 noma was identified in 3.1% (n=181; CI 2.6 to 3.8), stage 1 in 0.1% (n=10; CI 0.1 to 0.3) and stage 2 in 0.05% (n=3; CI 0.02 to 0.2). No children with stages 3–5 noma were detected in our study population (table 2). Based on these results, 3300 out of every 100 000 children in the group aged 0–15 years would have any stage of noma and 150 out of every 100 000 children would have stage 1 or 2 noma in the study area.

The prevalence of SAM in children aged 6 months to 5 years (n=3993) was 3.7% (n=149; CI 3.2 to 4.4) and MAM 7.7% (n=309; CI 6.7 to 8.7) (table 2).

Factors associated with noma

Table 3 describes univariable analysis of risk factors for stage 1 and 2 noma regardless of age category. This analysis showed that having eaten pap in the last 24 hours (OR 0.2; CI 0.1 to 0.9); the child eating pap once or more per week (OR 0.4; CI 0.1 to 0.9) and the caretaker preparing pap once or more per week compared with less frequent preparation of pap (OR 0.3; CI 0.1 to 0.8) were associated with not having stage 1 and 2 noma. The child experiencing an illness in the 12 months prior to the interview was associated with being a stage 1 or 2 noma case (OR 8.8; CI 1.1 to 69.5) (table 3).

The risk factors associated with any stage of noma for the group aged 0–5 years are shown in table 4. The multivariable analysis shows that two factors remained associated with being a noma case in the group aged 0–5 years, namely, having a well as the source of drinking water (adjusted odds ratio (aOR) 2.09; CI 1.22 to 3.60) and being aged 3–5 years (aOR 3.90; CI 2.04 to 7.47) (table 4).

In the group aged 6–15 years, the risk factors associated with any stage of noma are shown in table 5. Multivariable analysis showed that males were more likely to be noma cases (aOR 1.52; CI 1.04 to 2.22), and that the caretaker preparing pap once or more per week was associated with not having noma (aOR 0.36; CI 0.16 to 0.82) in the group aged 6–15 years (table 5).

Vaccination coverage rates in both age groups were low (21% of 0–5 year olds and 12% of 6–15 year olds had any immunisations noted on the vaccination card seen by the interviewer). No association between vaccination status and noma was seen in our study.

DISCUSSION

We have shown that the prevalence of any stage of noma in Kebbi and Sokoto States is 3.3%. Based on the study results, we therefore estimate that 129 120 (CI 105 294 to 152 947) individuals <15 years of age would have any stage of noma at the time of the survey within the two states. Most of these cases (n=120 082, 94% of all cases) would be children with simple gingivitis (ie, stage 0) and approximately 7101 (4% of all cases) and 1937 (2% of all cases) of children would have stage 1 and 2 noma, respectively. Our estimates exceeded those from Bello *et al* 2010–2018 period prevalence estimates (1.6 per 100 000)[11] and Fieger *et al.* in 2003 (6.4 per 1000 children).[10] Differences between the estimates could be due to geographical differences (Bello *et al* is north central Nigeria vs our northwest Nigeria), or due to methodological differences (Bello *et al* used patient record review of patients presenting at hospital and Fieger *et al* based their estimates on the number of clefts and mathematical modelling vs our community-based cross-sectional survey). It was unclear which stages of noma were included in these estimates.

Despite only covering two states of one country, our prevalence estimates would account for 17% of the current global WHO prevalence estimates.[7] Even though direct comparisons between the WHO and current study estimates are difficult as the stages included in the WHO estimates were not reported, our findings do suggest that the true burden of noma worldwide may be higher than previously thought.

Results from this study highlight the under-reported and overlooked nature of noma. Even though oral diseases, such as noma, are largely preventable, they impact over 3.5 billion people worldwide (untreated dental caries are the most prevalent of these oral health issues), disproportionally affecting marginalised groups.[25] Oral diseases are frequently more neglected than other diseases in low-income and middle-income countries, which may be linked to the fact that modern dentistry focuses on high-technology solutions, which are unaffordable and not currently feasible in low-resource settings.[26] This overarching neglect of oral diseases is magnified in the case of noma, as patients live in underserved, often rural locations.[27] Many cases will never seek care, and, even if they do, noma is unknown to many healthcare workers in endemic areas.[28] The condition may thus go undiagnosed, and rapid detection with opportunities for early treatment through improved oral hygiene, nutritional support and antibiotics, may be missed.

Strong surveillance systems have been the cornerstone of many successful neglected tropical disease control

Table 1 Demographic characteristics of households and children in the noma prevalence survey population

Households	n=3499 (%)
Caretaker age, years median (IQR)	30 (25–35)
Caretaker sex	
Female	3423 (97.8%)
Male	76 (2.2%)
Caretaker income source	
Employed or self-employed	2194 (30.8%)
Unemployed or other*	4927 (69.2%)
Total household members median (IQR)	5 (4–7)
Drinking water source	
Bore hole in the village	644 (18.4%)
River	91 (2.6%)
Tap (running water)	578 (16.2%)
Well in the compound	1512 (43.2%)
Other	674 (19.3%)
Treat water before drinking (yes)	1026 (29.3%)
Type of sanitation facility	
Flushing toilet	224 (6.4%)
Pit latrine (with slab)	650 (18.6%)
Pit latrine (no slab)	1168 (33.4%)
Other†	1457 (41.6%)

Children	Total n=7122 (%)‡	0–5 year olds n n=4239 (%)	6–15 year olds n n=2841 (%)
Age groups (years)			
0–5	4239 (59.8%)		
6–15	2841 (40.1%)		
Missing	42		
State			
Kebbi	3291 (46.5%)	2045 (48.2%)	1246 (43.9%)
Sokoto	3789 (53.5%)	2194 (51.8%)	1595 (56.1%)
Missing	42	0	0
Child sex			
Female	3692 (52.1%)	2119 (49.9%)	1573 (55.4%)
Male	3388 (47.9%)	2120 (50.0%)	1268 (44.6%)
Missing	42	0	0
Education of child			
None	5875 (83.0%)	3850 (90.8%)	2025 (71.3%)
Any education	1204 (17.0%)	388 (9.2%)	816 (28.7%)
Missing	42	1	0
Primary caretaker of the child interviewed			
Mother	6686 (94.4%)	4061 (95.8%)	2625 (92.4%)
Other (father, grandmother, grandfather)	394 (5.6%)	178 (4.2%)	216 (7.6%)
Missing	42	0	0

*Other caretaker income source includes being a housewife or student.
†Other sanitation facility includes neighbours house, the bush, river.
‡n=42 missing age category.

programmes.[29][30] The WHO has stated that robust surveillance helps to better understand the burden and distribution of disease, and to identify high-risk populations so that evidence-based decision-making can be used to target interventions in resource-constrained contexts.[31] A further benefit of robust surveillance is an increase in

Table 2 Noma stage and malnutrition prevalence (overall and by age group)

	Total (n=7122)*			0–5year olds n=4239 *			6–15year olds n=2841 *			P value (age comparison)
	n (%)	CI	DEFF	n (%)	CI	DEFF	n (%)	CI	DEFF	
Any noma	194 (3.3%)	2.7 to 4.0	1.9	63 (1.5%)	1.1 to 2.0	1.3	129 (4.4%)	3.6 to 5.4	1.6	<0.001
Noma stages										
None	6928 (96.6%)	95.9 to 97.2	1.9	4176 (98.5%)	98.0 to 98.9	1.3	2712 (95.6%)	94.6 to 96.4	1.6	<0.001
Stage 0: Simple gingivitis	181 (3.1%)	2.6 to 3.8	1.8	56 (1.3%)	1.0 to 1.8	1.3	123 (4.2%)	3.4 to 5.2	1.5	
Stage 1: Acute necrotising gingivitis	10 (0.1%)	0.1 to 0.3	1.2	6 (0.1%)	0.07 to 0.3	1	4 (0.1%)	0.04 to 0.4	1.5	
Stage 2: Oedema	3 (0.05%)	0.02 to 0.2	1	1 (0.02%)	0.0 to 0.2	1	2 (0.1%)	0.02 to 0.3	1	
Stage 3: Gangrene	0 (0%)	NA	NA	0 (0%)	NA	NA	0 (0%)	NA	NA	
Stage 4: Scarring	0 (0%)	NA	NA	0 (0%)	NA	NA	0 (0%)	NA	NA	
Stage 5: Sequela	0 (0%)	NA	NA	0 (0%)	NA	NA	0 (0%)	NA	NA	
Malnutrition				**6month–5year olds (n=3993)**						
Moderate acute malnutrition	NA	NA	NA	309 (7.7%)	6.7% to 8.7%	1.5				
Severe acute malnutrition	NA	NA	NA	149 (3.7%)	3.2% to 4.4%	1				

P value comparing age groups.
*44 missing age category
DEFF, design effect.

Table 3 Univariable analysis for stage 1 and 2 noma

	Study population		Univariable analysis		
	Proportion of all respondents; n=7122; %(n/N*)	Proportion respondents with stage 1 and 2 noma; n=13; % (n/N*)	OR	CI	P value
Primary caretaker					
Other	5.6% (394/7080)	7.7% (1/13)		Ref	0.561
Mother	94.4% (6686/7080)	92.3% (12/13)	0.5	0.07 to 4.18	
Pap eaten in the 24 hours before interview					
No	32.1% (2271/7080)	53.8% (7/13)		Ref	0.030
Yes	67.9% (4809/7080)	46.2% (6/13)	0.2	0.07 to 0.87	
Frequency of the child eating pap per week					
<1 or never	30.4% (2151/7080)	46.2% (6/13)		Ref	0.049
one or more	69.6% (4929/7080)	53.8% (7/13)	0.4	0.13 to 0.99	
Frequency of the caretaker preparing pap per week					
<1 or never	29.9% (2116/7080)	53.8% (7/13)		Ref	0.018
one or more	70.1% (4964/7080)	46.2% (6/13)	0.3	0.11 to 0.81	
Duration of breastfeeding at time of interview (months)					
12+	89.4% (6310/7061)	84.6% (11/13)		Ref	0.782
0–12	10.6% (751/7061)	15.4% (2/13)	1.2	0.27 to 5.63	
Colostrum given to the child at birth					
No	12.0% (843/7047)	15.4% (2/13)		Ref	0.366
Yes	88.0% (6204/7047)	84.6% (11/13)	0.5	0.10 to 2.32	
Child sick during last 12 months					
No	30.0% (2,131/7,080)	7.7% (1/13)		Ref	0.041
Yes	70.0% (4949/7080)	92.3% (12/13)	8.8	1.11 to 69.49	
Did you seek healthcare for this child in the last year?					
No	48.4% (3428/7080)	23.1% (3/13)		Ref	0.221
Yes	51.6% (3652/7080)	76.9% (10/13)	2.5	0.58 to 10.51	
Vegetables eaten in the 24 hours before interview					
No	68.2% (4829/7080)	76.9% (10/13)		Ref	0.461
Yes	31.8% (2251/7080)	23.1% (3/13)	0.6	0.13 to 2.50	
Wealth score (mobile phone, motorbike, tractor, camel)					
0–1	63.5% (4522/7122)	84.6% (11/13)		Ref	0.106
2–4	36.5% (2600/7122)	15.4% (2/13)	0.3	0.08 to 1.27	

Analysis adjusted for the survey design.
P value from logistic regression model.
*n=total number of respondents who answered the question (excluding missing).

the number of cases identified, diagnosed and treated.[32] Due to the neglected nature of noma, surveillance activities for active noma cases are hampered and it is unlikely that current surveillance mechanisms adequately identify deaths from noma at a community level. The mortality rate associated with noma is unknown, but estimated to be as high as 90% if the disease is left untreated.[1] Deaths may be primarily due to starvation, aspiration pneumonia, respiratory insufficiency or sepsis,[33][34] and not be attributed to noma, further reducing the potential for accurate reporting of disease burden. Our findings

suggest that improved efforts to enumerate the burden of disease are necessary.

This study highlights the need for a single classification system for the differential diagnosis of each stage of noma, which would be beneficial in standardising reporting of noma globally by the clinical and research noma community. In published work, noma is often classified into two stages (acute and chronic noma[35][36]) or with the Montandon system (classifies noma according to the location of the defect).[37] The lack of standardisation complicates comparison between different studies. The

Table 4 Univariable and multivariable analyses of associations with any noma cases (stage 0 to 5), 0–5 years

	Study population		Univariable analysis			Multivariable analysis		
	Proportion of all respondents; n=4239; %(n/N*)	Proportion respondents with any noma stage; n=63; % (n/N*)	OR	CI	P value	aOR	CI	P value
Child demographics								
Child age (years)								
0–2	46.2% (1957/4239)	17.5% (11/63)		Ref	<0.001		Ref	<0.001
3–5	53.8% (2282/4239)	82.5% (52/63)	4.1	2.20 to 7.62		3.9	2.04 to 7.47	
Birth order								
1–2	39.9% (1691/4239)	30.2% (19/63)		Ref	0.174		Ref	0.398
3 or more	60.1% (2548/4239)	69.8% (44/63)	1.5	0.83 to 2.88		1.36	0.67 to 2.79	
Feeding practices								
Duration of breastfeeding								
12+ months	84.4% (3565/4226)	95.2% (60/63)		Ref	0.02			
0–12 months	15.6% (661/4226)	4.8% (3/63)	0.3	0.09 to 0.81				
Colostrum given to baby								
No	11.9% (502/4221)	11.1% (7/63)		Ref	0.899			
Yes	88.1% (3719/4221)	88.9% (56/63)	1.1	0.37 to 3.09				
Frequency of the child eating pap per week								
<1 or never	31.6% (1340/4239)	34.9% (22/63)		Ref	0.597			
1 or more	68.4% (2899/4239)	65.1% (41/63)	0.9	0.47 to 1.54				
Frequency of the caretaker preparing pap per week								
<1 or never	30.8% (1307/4239)	34.9% (22/63)		Ref	0.505			
1 or more	69.2% (2932/4239)	65.1% (41/63)	0.8	0.46 to 1.46				
Animal products eaten in the 24 hours before interview								
No	91.5% (3879/4239)	95.2% (60/63)		Ref	0.274			
Yes	8.5% (360/4239)	4.8% (3/63)	0.5	0.16 to 1.67				
Grains eaten in the 24 hours before interview								
No	19.3% (819/4239)	22.2% (14/63)		Ref	0.607			
Yes	80.7% (3420/4239)	77.8% (49/63)	0.8	0.45 to 1.59				
Vegetables eaten in the 24 hours before interview								
No	69.6% (2952/4239)	69.8% (44/63)		Ref	0.963			
Yes	30.4% (1287/4239)	30.2% (19/63)	1	0.56 to 1.73				
Health								
Are the teeth ever cleaned (self-reported)								
No	13.8% (508/3679)	9.7% (6/62)		Ref	0.3			
Yes	86.2% (3171/3679)	90.3% (56/62)	1.5	0.69 to 3.39				
Teeth cleaning frequency per day (self-reported)								
Once or twice	85.1% (3132/3679)	88.7% (55/62)		Ref	0.37			
Less than once	14.9% (547/3679)	11.3% (7/62)	0.7	0.34 to 1.50				
SAM, MAM								
Normal	88.5% (3535/3993)	91.1% (51/56)		Ref	0.199			
SAM	3.7% (149/3993)	7.1% (4/56)	1	0.55 to 6.85				
MAM	7.7% (309/3993)	1.8% (1/56)	0.2	0.03 to 1.71				
GAM								
Normal	88.5% (3535/3993)	91.1% (51/56)		Ref	0.666			
GAM	11.5% (458/3993)	8.9% (5/56)	0.8	0.26 to 2.36				

Continued

Table 4 Continued

	Study population		Univariable analysis			Multivariable analysis		
	Proportion of all respondents; n=4239; %(n/N*)	Proportion respondents with any noma stage; n=63; % (n/N*)	OR	CI	P value	aOR	CI	P value
Was the child vaccinated (self-report)								
No	27.5% (1165/4239)	23.8% (15/63)		Ref	0.516			
Yes	72.5% (3074/4239)	76.2% (48/63)	1.2	0.67 to 2.20				
Vaccinations listed on vaccination card								
Diphtheria								
No	71.1% (635/893)	75.0% (12/16)		Ref	0.688			
Yes	28.9% (258/893)	25.0% (4/16)	0.8	0.21 to 2.81				
Pertussis								
No	73.7% (658/893)	75.0% (12/16)		Ref	0.84			
Yes	26.3% (235/893)	25.0% (4/16)	0.9	0.24 to 3.21				
Tetanus								
No	82.1% (733/893)	75.0% (12/16)		Ref	0.584			
Yes	17.9% (160/893)	25.0% (4/16)	1.4	0.40 to 5.11				
Hepatitis A								
No	93.4% (834/893)	93.8% (15/16)		Ref	0.9			
Yes	6.6% (59/893)	6.3% (1/16)	0.9	0.11 to 6.86				
Hepatitis B								
No	77.4% (691/893)	68.8% (11/16)		Ref	0.415			
Yes	22.6% (202/893)	31.3% (5/16)	1.6	0.52 to 4.84				
Measles								
No	38.0% (339/893)	31.3% (5/16)		Ref	0.628			
Yes	62.0% (554/893)	68.8% (11/16)	1.3	0.43 to 4.11				
Pneumococcal disease								
No	69.4% (620/893)	75.0% (12/16)		Ref	0.686			
Yes	30.6% (273/893)	25.0% (4/16)	0.8	0.19 to 2.96				
Yellow fever								
No	64.7% (578/893)	68.8% (11/16)		Ref	0.745			
Yes	35.3% (315/893)	31.3% (5/16)	0.8	0.24 to 2.75				
Meningitis								
No	87.6% (782/893)	75.0% (12/16)		Ref	0.15			
Yes	12.4% (111/893)	25.0% (4/16)	2.3	0.74 to 7.26				
Polio								
No	21.8% (195/893)	12.5% (2/16)		Ref	0.351			
Yes	78.2% (698/893)	87.5% (14/16)	2	0.48 to 8.12				
Any vaccination listed on vaccination card								
No	79.2% (3356/4239)	74.6% (47/63)		Ref	0.415			
Yes	20.8% (883/4239)	25.4% (16/63)	1.3	0.70 to 2.40				
Child sick during last 12 months								
No	29.7% (1260/4239)	23.8% (15/63)		Ref	0.235			
Yes	70.3% (2979/4239)	76.2% (48/63)	1.4	0.82 to 2.28				
How often child was sick, last 12 months								
0–1	50.9% (2156/4239)	50.8% (32/63)		Ref	0.994			
2 or more	49.1% (2083/4239)	49.2% (31/63)	1	0.60 to 1.68				

Table 4 Continued

	Study population		Univariable analysis			Multivariable analysis		
	Proportion of all respondents; n=4239; %(n/N*)	Proportion respondents with any noma stage; n=63; % (n/N*)	OR	CI	P value	aOR	CI	P value
Did you seek healthcare for this child in the last year?								
No	47.3% (2006/4239)	50.8% (32/63)		Ref	0.634			
Yes	52.7% (2233/4239)	49.2% (31/63)	0.9	0.50 to 1.53				
Difficulties accessing healthcare (cost, time, distance)								
Didn't seek care	47.3% (2006/4239)	50.8% (32/63)		Ref	0.765			
No difficulties	46.0% (1949/4239)	44.4% (28/63)	0.9	0.51 to 1.60				
Yes, there were difficulties	6.7% (284/4239)	4.8% (3/63)	0.7	0.20 to 2.08				
Caretaker and household information								
Caretaker age (years)								
Under 30	49.8% (2109/4239)	34.9% (22/63)		Ref	0.01		Ref	0.251
30 or older	50.2% (2130/4239)	65.1% (41/63)	1.8	1.16 to 2.94		1.38	0.80 to 2.39	
Primary caretaker of the child interviewed								
Other	4.2% (178/4239)	7.9% (5/63)		Ref	0.166			
Mother	95.8% (4061/4239)	92.1% (58/63)	0.5	0.18 to 1.34				
Total number of household members								
0–6	71.7% (3039/4239)	74.6% (47/63)		Ref	0.591			
Above 6	28.3% (1200/4239)	25.4% (16/63)	0.9	0.48 to 1.51				
Drinking water source								
Other (borehole, river, tap)	54.9% (2326/4239)	36.5% (23/63)		Ref	0.007		Ref	0.008
Well	45.1% (1913/4239)	63.5% (40/63)	2.1	1.24 to 3.66		2.09	1.22 to 3.60	
Water treatment								
No	71.2% (3020/4239)	73.0% (46/63)		Ref	0.759			
Yes (strain through cloth, let stand and settle, boil)	28.8% (1219/4239)	27.0% (17/63)	0.9	0.54 to 1.57				
Wealth score (mobile phone, motorbike, tractor, camel)								
0–1	63.5% (2693/4239)	65.1% (41/63)		Ref	0.81			
2–4	36.5% (1546/4239)	34.9% (22/63)	0.9	0.55 to 1.60				

Analysis adjusted for the survey design.
Variables with 10 or more cases and a p<0.2 in the univariable analysis included in the multivariable model (child age, birth order, caretaker age, drinking water source).
P value from logistic regression model.
aOR, adjusted odds ratio.

WHO noma staging system[1] is the most comprehensive to date and includes an early-stage noma definition which is useful as it identifies those at risk of progressing to later stage noma. However, it lacks specificity as it overlaps with commonly seen ailments such as simple gingivitis and acute necrotising gingivitis and therefore may overestimate the burden of disease. It is currently unknown what the risk factors are for progression of noma, nor the proportion of patients who progress to the later stages of disease. Explicit reference to which WHO stages of noma are included in prevalence and incidence estimates as well as improved detail of the method employed in these estimations would greatly improve our ability to compare findings across studies. The lack of consistency of approach to assessing the incidence and prevalence of noma in the literature and the lack of real investment in assessing the true incidence and prevalence of this condition particularly in regions that bear the highest global burden contribute to the ongoing neglect of this disease and the populations it affects.

Study findings indicate that children aged between 3 and 5 years had a higher prevalence of noma in comparison to those aged 2 years or less, a finding corroborated by other studies.[38–41] We hypothesise that this finding is likely due to the relationship between child feeding practices in Nigeria and malnutrition as a risk factor for noma. Our study did not identify an association between acute malnutrition and having noma. However, other

Table 5 Univariable and multivariable analyses of associations with any noma cases (stage 0 to 5), 6–15 years

	Study population		Univariable analysis			Multivariable analysis		
	Proportion of all respondents; n=2841; %(n/N*)	Proportion respondents with any noma stage; n=129; % (n/N*)	OR	CI	P value	aOR	CI	P value
Demographics								
Child age (years)								
6–10	74.9% (2127/2841)	76.0% (98/129)		Ref	0.722			
11–15	25.1% (714/2841)	24.0% (31/129)	0.93	0.64 to 1.36				
Child gender								
Female	55.4% (1573/2841)	45.7% (59/129)		Ref	0.036		Ref	0.031
Male	44.6% (1268/2841)	54.3% (70/129)	1.5	1.03 to 2.20		1.52	1.04 to 2.22	
Birth order								
1–2	50.2% (1426/2841)	55.8% (72/129)		Ref	0.186		Ref	0.613
3 or more	49.8% (1415/2841)	44.2% (57/129)	0.79	0.56 to 1.12		0.9	0.61 to 1.34	
Feeding practices								
Colostrum given to baby								
No	12.1% (341/2826)	11.7% (15/128)		Ref	0.856			
Yes	87.9% (2485/2826)	88.3% (113/128)	1.06	0.57 to 1.98				
Frequency of the child eating pap per week								
<1 or never	28.5% (811/2841)	36.4% (47/129)		Ref	0.042		Ref	0.136
1 or more	71.5% (2030/2841)	63.6% (82/129)	0.68	0.47 to 0.98		1.87	0.83 to 4.21	
Frequency of the caretaker preparing pap per week								
<1 or never	28.5% (809/2841)	38.8% (50/129)		Ref	0.011		Ref	0.015
1 or more	71.5% (2032/2841)	61.2% (79/129)	0.61	0.42 to 0.89		0.36	0.16 to 0.82	
Animal products eaten in the 24 hours before the interview								
No	91.0% (2585/2841)	94.6% (122/129)		Ref	0.265			
Yes	9.0% (256/2841)	5.4% (7/129)	0.57	0.22 to 1.52				
Grains eaten in the 24 hours before the interview								
No	18.2% (516/2841)	15.5% (20/129)		Ref	0.455			
Yes	81.8% (2325/2841)	84.5% (109/129)	1.22	0.73 to 2.04				
Vegetables eaten in the 24 hours before the interview								
No	66.1% (1877/2841)	71.3% (92/129)		Ref	0.293			
Yes	33.9% (964/2841)	28.7% (37/129)	0.78	0.50 to 1.23				
Health								
Are the teeth ever cleaned (self-reported)								
No	3.1% (88/2823)	4.7% (6/128)		Ref	0.372			
Yes	96.9% (2735/2823)	95.3% (122/128)	0.63	0.23 to 1.73				
Teeth cleaning method (self-reported)								
Toothbrush	23.8% (677/2841)	18.6% (24/129)		Ref	0.654			
Ash or cloth	1.9% (55/2841)	2.3% (3/129)	1.54	0.45 to 5.32				
Salt and water or stick	16.4% (466/2841)	15.5% (20/129)	1.26	0.68 to 2.32				
None or other	57.8% (1643/2841)	63.6% (82/129)	1.44	0.82 to 2.52				
Teeth cleaning frequency per day (self-reported)								
Once or twice	95.2% (2687/2823)	93.0% (119/128)		Ref	0.275			
Less than once	4.8% (136/2823)	7.0% (9/128)	1.53	0.72 to 3.26				
Any vaccinations listed on vaccination card								
No	88.5% (2513/2841)	86.0% (111/129)		Ref	0.451			
Yes	11.5% (328/2841)	14.0% (18/129)	1.24	0.71 to 2.17				

Continued

Table 5 Continued

	Study population		Univariable analysis			Multivariable analysis		
	Proportion of all respondents; n=2841; %(n/N*)	**Proportion respondents with any noma stage; n=129; % (n/N*)**	**OR**	**CI**	**P value**	**aOR**	**CI**	**P value**
Polio vaccination (self-report)								
No	29.8% (846/2841)	24.0% (31/129)		Ref	0.131		Ref	0.113
Yes	70.2% (1995/2841)	76.0% (98/129)	1.36	0.91 to 2.01		1.4	0.92 to 2.13	
Child sick last 12 months								
No	30.7% (871/2841)	24.0% (31/129)		Ref	0.148		Ref	0.138
Yes	69.3% (1970/2841)	76.0% (98/129)	1.39	0.89 to 2.18		1.51	0.88 to 2.60	
How many times was child sick during last 12 months								
0–1	51.1% (1451/2841)	46.5% (60/129)		Ref	0.379			
2 or more	48.9% (1390/2841)	53.5% (69/129)	1.19	0.81 to 1.74				
Did you seek healthcare for this child in the last year?								
No	50.1% (1422/2841)	47.3% (61/129)		Ref	0.62			
Yes	49.9% (1419/2841)	52.7% (68/129)	1.1	0.75 to 1.63				
Difficulties accessing healthcare (cost, time, distance)								
Didn't seek care	50.1% (1422/2841)	47.3% (61/129)		Ref	0.068		Ref	0.259
No difficulties	43.9% (1247/2841)	41.9% (54/129)	0.99	0.63 to 1.55		0.78	0.45 to 1.36	
Yes, there were difficulties	6.1% (172/2841)	10.9% (14/129)	1.98	1.12 to 3.51		1.44	0.73 to 2.85	
Caretaker and household information								
Caretaker age (years)								
Under 30	21.5% (611/2841)	20.2% (26/129)		Ref	0.731			
30 or older	78.5% (2230/2841)	79.8% (103/129)	1.08	0.69 to 1.69				
Caretaker gender								
Female	97.5% (2770/2841)	99.2% (128/129)		Ref	0.271			
Male	2.5% (71/2841)	0.8% (1/129)	0.32	0.04 to 2.40				
Primary caretaker of the child interviewed								
Other	7.6% (216/2841)	8.5% (11/129)		Ref	0.712			
Mother	92.4% (2625/2841)	91.5% (118/129)	0.87	0.43 to 1.78				
Total number of household members								
0–6	58.1% (1650/2841)	67.4% (87/129)		Ref	0.025		Ref	0.145
Above 6	41.9% (1191/2841)	32.6% (42/129)	0.66	0.46 to 0.95		0.74	0.49 to 1.11	
Drinking water source								
Other (borehole, river, tap)	57.8% (1641/2841)	54.3% (70/129)		Ref	0.498			
Well	42.2% (1200/2841)	45.7% (59/129)	1.16	0.79 to 1.87				
Water treatment								
No	69.1% (1964/2841)	65.1% (84/129)		Ref	0.376			
Yes (strain through cloth, let stand and settle, boil)	30.9% (877/2841)	34.9% (45/129)	1.21	0.79 to 1.87				
Wealth score (mobile phone, motorbike, tractor, camel)								
0–1	62.9% (1787/2841)	70.5% (91/129)		Ref	0.063		Ref	0.19
2–4	37.1% (1054/2841)	29.5% (38/129)	0.7	0.48 to 1.02		0.77	0.52 to 1.14	

Analysis adjusted for the survey design.
Variables with 10 or more cases and a p<0.2 in the univariable analysis included in the multivariable model (child gender, birth order, frequency of the child eating pap per week, frequency of the caretaker preparing pap per week, polio vaccination, child sick last 12 months, difficulties accessing health carehealthcare, total number of household members, wealth score.
P value from logistic regression model.
aOR, adjusted odds ratio; p, p value from logistic regression model.

studies have shown that rural Nigerian children typically breastfeed until 24 months of age[42] and then transition to a limited diet.[43] This has shown to result in higher levels of malnutrition and stunting[43] and therefore, a potentially higher risk of developing noma. This discrepancy in our findings in comparison to other studies could be due to the fact that our population had early stage noma, whereas other studies could have identified the association between malnutrition and late stage noma. It is also possible that late stage noma could cause malnutrition if the child was having difficulties eating due to the noma infection.

Our findings showed that older (aged 6–15 years) male children were more likely to have noma in comparison to older female children. We do not know what would explain this finding as we would not expect an inherent difference in gender in noma development. This finding warrants further research.

We showed that various factors relating to pap preparation and consumption were linked to not having stage 1 and 2 noma among the whole study sample and of any stage noma in the group aged 6–15 years old. It is difficult to explain this finding as it could be due to a lack of access to food or that pap made more frequently has less chance of becoming contaminated (thus causing less disease) as it is stored for shorter periods of time. A Nigerian study showed that when mothers prepared food far in advance, contamination was more likely to occur.[44] A further Nigerian study in Kebbi State showed that pap was contaminated with high levels of Salmonella in comparison to other commonly eaten foods.[45] This finding affected children in the older age group which may be because they are more reliant on this food source in comparison to the younger children.

This study further indicated that having a well as the main water source in comparison to other water sources such as a borehole, river or tap, was associated with having noma in the group aged 0–5 years. Well water has a high risk of contamination from nearby pit latrines or livestock,[46] and the consumption of contaminated well water is a risk factor for diarrhoea,[47] which in turn is an identified comorbidity for children with noma.[48]

Vaccination coverage in all eligible children included in the study was low. Even though this result prevented us from exploring whether vaccination is associated with noma prevention, it does confirm findings from other studies in rural Sokoto State, where up to 70% of children were not vaccinated against measles and other common childhood diseases.[35] Low immunisation coverage is an important indicator of struggling societal systems in need of multisectoral improvements, including access to quality timely healthcare, access to safe drinking water, improved nutrition and security. Prevention efforts should also include early detection training with healthcare workers, and setting up effective referral pathways. These initiatives are resource-intensive and require large-scale investment of time, money and human resources.

This study had a large sample size and robust approach to sampling and analysis, and we are confident that prevalence estimates are broadly representative of the study area. However, a few limitations should be considered. This cross-sectional study was conducted on a disease with an extremely rapid clinical progression with onset to death taking as little as 2 weeks.[1] Thus, it is possible that Neyman bias was present and we only identified a fraction of noma cases that occur. The research team did come across stage five noma patients in study villages, but not in households included in the study. These patients were referred to the NCH for care. It is possible that a study with a larger sample size could have identified children with the later stages of disease. Some of the answers were self-reported by caretakers, which could have introduced social desirability bias that either inflated or deflated the risk factor associations found in the study. This aspect was mitigated by anonymising the survey and trying to phrase questions in contextually acceptable ways. Data on some risk factors, such as comorbidities, water quality and malnutrition in older children were not collected, limiting our ability to identify associations with these factors. It is difficult to interpret the results of questions asking about consumption of any food and specific foodstuffs in the 24 hours prior to the interview and associations with noma as respondents with noma could have difficulty eating in general and would thus have been less likely to report eating at all. Finally, the challenging security situation limited the areas the research teams could access. This may have introduced selection bias, and an underestimation of noma and malnutrition prevalence, as we did not visit the hardest to reach communities who were likely most vulnerable to noma infection. Future research on the burden of noma should be combined with existing surveillance systems for other disease and research activities such as malnutrition and vaccination surveys.

Noma meets the criteria of a neglected tropical disease as defined by the WHO: it is a preventable disease that affects children in low-resource contexts; children that survive will have life-long physical and mental health sequelae; and there is poor understanding of the disease, its pathogenesis and global burden.[49] This study has shown that the prevalence of any stage of noma is higher than previous estimates. While we did not find any later stages of the disease, the high rates of simple gingivitis and the presence of known risk factors for noma (low vaccination rates, malnutrition and poor access to healthcare) suggest the need for improved coverage of preventative interventions and access to care in northwest Nigeria. Our prevalence estimates are greater than those for snakebite in Nigeria (497 per 100 000 people), which the WHO recently recognised as a neglected tropical disease.[50] Noma prevention and control will require a concerted health systems approach. Adding noma to WHO's list of neglected tropical diseases will facilitate global attention for noma and the allocation of much-needed resources to those countries where noma continues to be a public health problem.

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