


Research Article

An Update on the Epidemiology of Noma (Facial Gangrene) in Ethiopia

Heron Gezahegn Gebretsadik

Abstract

Noma is an infectious disease that mainly affects debilitated children in developing countries. The geographic region of the noma belt, stretching from Senegal to Ethiopia, is arguably the most affected area in the world. This updated retrospective cross-sectional study assessed the epidemiology of Noma in Ethiopia. The assessment was based on a review of patients' medical records from three noma treatment centers. A modified case report form (CRF) was used to collect the clinical information required for data analysis. One hundred eighty-two patient records from the medical registries of the three centers were analyzed to determine the results of the updated study. Data analysis revealed that the disease occurred primarily in children younger than ten years of age. However, the condition also occurred in neonates, infants, children, and adults. Amhara and Oromia regions had a high number of noma cases, 29.2% (n=38) and 25.4% (n=33), respectively. The geographical distribution also suggests that noma is a tropical disease. Cheeks, lower lips, and upper lips were the most commonly affected orofacial anatomic regions. Indeed, more attention should be paid to these geographic regions and children younger than ten.

Keywords: Epidemiology, Noma, Cancrum oris, Ethiopia, Cross-sectional study

Abbreviations

GBD: Global Burden of Disease

NTDs: Neglected Tropical Diseases

CRF: Case Report Form

WHO: World Health Organization

DALYs: Life years adjusted for disabilities

ANUG: Acute necrotizing ulcerative gingivitis

AIDS: Acquired Immunodeficiency Syndrome

Introduction

Noma is reported to be an old companion of humankind [1]. However, the disease has disappeared in developed countries due to gradual socioeconomic progress [2]. Today, Noma is most prevalent in the world's most disadvantaged populations [3]. Some believe that the Noma was first described by Hippocrates in the 5th century BC when he described a destructive ulceronecrotic lesion on a patient's face, mouth, and respiratory tract [4]. In the late Middle Ages, "water canker" and "mouth canker" were some of the many names for Noma, and the Latin name "cancrum oris" still in use today emerged, although

Affiliation:

Euclid University, School of Global Health and Bioethics, The Gambia

*Corresponding author:

Heron Gezahegn Gebretsadik, Euclid University, School of Global Health and Bioethics, The Gambia

Citation: Heron Gezahegn Gebretsadik. An Update on the Risk Factors of Noma (Facial Gangrene) in Ethiopia. Fortune Journal of Health Sciences 6 (2023): 109-114.

Received: March 08, 2023

Accepted: March 14, 2023

Published: March 24, 2023

contemporary physicians already recognized the difference between Noma and cancer [5]. Between the 16th and 18th centuries AD, reports of Noma became more frequent and precise throughout Europe, and the usual conservative treatment of simple rinses and pulling loose teeth was increasingly supplemented by newly developed surgical methods [2]. In the wake of improved hygiene, socioeconomic status, and nutritional conditions in much of the increasingly industrialized societies in the 19th and 20th centuries AD, Noma almost completely disappeared in the more developed regions, except for notable recurrences during the two world wars and especially in prisoner-of-war and concentration camps and, more recently, in immunocompromised patients [6, 7]. From the historical excerpt, it is clear that Noma is not an NTD per se but rather a neglected disease [8]. Because of its pathology and close association with poverty, Noma has also become known as the "face of poverty" [9]. Over time, the World Health Organization (WHO) has classified Noma as an NTD, oral disease, and non-communicable disease. Currently, the disease is listed in the International Classification of Diseases version 10 (ICD-10) under "certain infectious and parasitic diseases" as "A69.0 Necrotizing ulcerative stomatitis". It is also referred to as cancrum oris, fusospirochaetal gangrene, Noma, and stomatitis gangrenous [10]. However, many different historical or local names still exist [11].

Today, most reported noma patients are children well under 10 years of age, but cases in adolescents and adults may occur sporadically [12]. Most cases live in sub-Saharan Africa's poorest and most remote regions [13]. Historically, Noma was most prevalent in the areas known as the Noma belt in Mauritania, Senegal, Mali, Niger, Chad, Sudan, and Ethiopia [14, 15]. Nowadays, the burden of disease studies is an integral part of descriptive epidemiology, with GBD studies becoming the most comprehensive descriptive epidemiological studies worldwide [16]. The results of the GBD studies are widely cited, and GBD core investigators are among the most influential scientists in the world" [17]. The most recent version, the 2015 GBD study, evaluated 315 different causes of health harm [16, 18]. However, the list of GBD causes is far from exhaustive, which can lead to distorted perceptions. Not explicitly listed causes do not appear to exist because they do not come with a precise price tag in terms of DALYs. As a result, such unlisted causes are often disregarded by the broader (global) public health community and stakeholders, adversely affecting resource allocation and efforts to prevent and address them. Not surprisingly, these problems are more pronounced in neglected diseases such as Noma, which primarily affect disadvantaged populations [19]. The present study aimed to address this suboptimal situation by examining the epidemiology of a neglected disease, namely Noma, in Ethiopia.

Materials and Methods

Research Design

A descriptive retrospective cross-sectional study was conducted to update the epidemiology of Noma in Ethiopia. Revision of patient charts obtained from the medical registry of Yekatit 12 Hospital, Facing Africa Ethiopia, and Harar Project Ethiopia was carried out to report the latest data on the epidemiology of Noma in Ethiopia.

Sample and setting

This updated cross-sectional study was conducted in Addis Ababa. Medical records of patients diagnosed with Noma between March 2004 and December 2022 were used for data analysis. Patient records were obtained from the three main noma treatment centers in Ethiopia.

Data collection and analysis

A modified case report form was used to collect the required data. The clinical and demographic data collected in the modified reporting forms were used to describe the epidemiology of Noma in Ethiopia. Data management and analysis began with registering clinical and demographic data in an Excel spreadsheet. The IBM SPSS (Statistical Package for Social Sciences) Statistics, version 29 version was used to analyze the collected data. Descriptive statistical analysis was applied to describe and present the findings of the study in simple and compact forms.

Ethical clearance

Ethical approval for the study was obtained from the Institutional Review Board (IRB) of the Department of Health in Addis Ababa.

Findings

Three newly admitted cases (admitted in 2021 and 2022) and 16 old cases that were not amenable to participation in the previous survey for various reasons were included in the updated review. The updated study included the data of 182 Noma cases for data analysis. Medical records of patients admitted to the three centers between March 2004 and January 2023 were used for the review. Accordingly, 182 medical records were obtained from Ethiopia's three major noma treatment centers. Of these, 81, 68, and 33 were obtained from Facing Africa Ethiopia, Yekatit-12 Hospital, and Harar Project Ethiopia, respectively.

Sex ratio of noma cases

There were more women (63.7%) than men (36.3%).

Age at the admission of Noma cases

The age at noma case admission ranged from 1 year to 70 years. Thus, 32.4% (n=59) were 11-20 years old and most common. This age group was found to be followed by 27.5% (n=50) and 15.9% (n=29) of 21-30 years old and 31-40 years

old, respectively. The 41-50, 1-10, 51-60, and 61-70 age groups accounted for 11% (n=20), 7.1% (n=13), 4.4% (n=8), and 1.7% (3), respectively, of the total noma cases studied (Table 1). In general, 92.9% of the medical records showed that the patients were admitted at the age of 11 and above.

Residential regions of the Noma cases

Of the total 130 (111 from the previous study) medical records indicating the place of birth and residence of noma cases, 28.5% (n=37) and 25.4% (n=33) were from the Amhara and Oromia regions, respectively. This is followed by Southern Nations, Nationalities People, and Somali regions with 13.1% (n=17) and 9.2% (n=12), respectively. The fewest noma cases were reported from the Harari regional state and Dire Dawa city administration at 1.5% (n=2) each (Table 2). Based on available data, the geographic distribution of Noma cases in Ethiopia was described as shown in Figure 1 below.

Noma case admission time in years

Of these 182 total medical records reviewed for date of admission, 44.5% (n=81), 39.6% (n=72), and 15.9% (n=29) were admitted between 2016-2022, 2010-2015, and 2004-2009, respectively.

Table 1: Age groups by years at the admission of noma cases

Age groups by years at admission	Quantity	Percentage
≤ 10	13	7.10%
11-20	59	32.40%
21-30	50	27.50%
31-40	29	15.90%
41-50	20	11%
51-60	8	4.40%
61-70	3	1.70%
Total	182	100%

Table 2: Reported number of Noma cases in Ethiopia

Regions and city administrations	Number of noma cases identified	Percentage
Addis Ababa	3	2.30%
Afar	6	4.60%
Amhara	37	28.50%
Dire Dawa	2	1.60%
Gambella	9	6.90%
Harari	2	1.60%
Oromia	33	25.40%
Somali	12	9.20%
SNNPR	17	13.10%
Tigray	9	6.90%
Total	130	100%

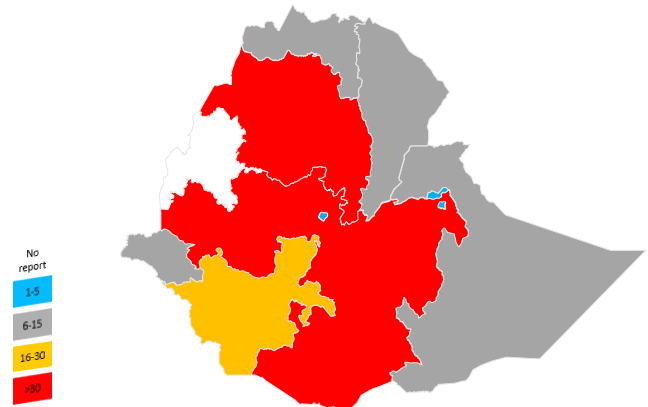


Figure 1: Reports of Noma cases in Ethiopia (2004-2022)

Date of Onset of Noma

Of these 123 (111 from the previous study) medical records reporting time of onset, 88.6% (n=109) reported onset of Noma between 1-9 years of age. A single case reported the onset of the disease since birth. In three other patients, the condition occurred in infancy. Four noma cases reported the onset of the disease at age ten years. The remaining six affected individuals reported the onset of the disease at ages 14, 16, 18, 22, 27, and 29 years.

Noma-induced defects of the face

From the medical records reviewed, the left side (n=95) of the face and mouth was more commonly affected by Noma than the right side (n=81). In general, the medical records reported either left-sided or right-sided anatomic defects in 96.7% (n=176). The remaining 3.3% (n=6) of medical records reported neither left-sided nor right-sided facial deformities. The most common left-, right-, and neither left- nor right-sided noma-induced defects are described in Table 2.

Discussion

Over the past 25 years, and particularly in the wake of the Global burden of disease (GBD) studies, quantifying health using disability-adjusted life-years (DALYs) has gained prominence [17]. DALYs and related measures of health-adjusted life years (e.g., quality-adjusted life years, or QALYs) aim to quantify health gains or losses due to premature death and nonfatal health outcomes in a single, continuous, proportionally scaled, and uniform measure [20] (see Chapter 3.2. for a formal definition of DALYs) (21). An explicit assumption is that such time-based health burden estimates allow direct comparison of health losses and gains across conditions, interventions, locations, populations, and time points. Combining information on mortality and morbidity into a single measure is of great importance because "death does not tell the whole story" [22] about health. Therefore, DALYs and QALYs are increasingly used for monitoring, evaluation, and resource allocation and have

Table 3: Distribution of noma-induced facial defects by anatomy.

Damaged anatomical region(s) of the face	Frequency of defects			
	Left side	Right side	Neither right nor left side (complete loss)	Total
Cheek	19	11	-	30
Lower lip	10	9	-	19
Upper lip	10	6	-	16
Cheek, lower and upper lip	4	6	-	10
Cheek, corner of the mouth, lower and upper lip	11	8	-	17
Cheek & lower lip	4	6	-	10
Cheek, nose, lower and upper lip	5	4	-	9
Cheek & upper lip	5	4	-	9
Lower and upper lips	4	2	-	6
Corner of the mouth, lower and upper lip	4	2	-	6
Nose	3	3	-	6
Nose and upper lip	3	3	-	6
Cheek, nose and upper lip	1	3	-	4
Corner of the mouth and upper lip	2	2	-	4
Nose and upper lip	3	3	-	6
Cheek and corner of the mouth	-	3	-	3
Oral commissure	2	1	-	3
Upper jaw and upper lip	1	2	-	3
Lower lip	-	-	2	2
Nose	-	-	2	2
Central lower lip	-	-	1	1
Central upper lip and nose	-	-	1	1
Chin and upper lip	1	-	-	1
Cheek & Nose	-	1	-	1
Cheek, lower lip, nose and lower jaw	1	-	-	1
Infraorbital area, corner of the mouth, nose and zygomatic bone	1	-	-	1
Hard palate, upper jaw, nose, upper lip and zygomatic bone	1	1	-	2
Total	95	81	6	182

= not identified

become an important "currency" in health care. Institutions with international (e.g., World Health Organization (WHO), World Bank, Bill & Melinda Gates Foundation) and national reach (e.g., National Institute for Health and Care Excellence (NICE) in the United Kingdom) in search of rational and evidence-based criteria against which to prioritize health investment decisions have been instrumental in the development and dissemination of DALYs and QALYs [17, 20].

In contrast, primary epidemiologic data for certain diseases such as Noma are lacking [23]. This is because neglected diseases such as Noma occur predominantly in poor communities that do not keep health records and often have nomadic lifestyles [24]. In addition, the disease is often

concealed by families who consider it a bad omen [25]. A recent report estimated an annual incidence of 25,000 cases in developing countries bordering the Sahara. However, these estimates reflect only the tip of the iceberg, as at most 10% of those affected seek medical attention [26]. Noma occurs primarily in children under six years of age in countries with a low human development index, with the highest incidence in the region known as the noma belt. This region spans West and East Africa, from Senegal to Ethiopia [27, 28]. A study in Senegal found an annual incidence rate of Noma in children ranging from 1.2 to 4.2 cases per million [29, 30]. On the other hand, a study conducted in northwestern Nigeria concluded that the incidence of Noma was estimated at 6.4 per 1000 children [31].

Further studies in Nigeria estimated the incidence to be between 1.6 and 8.3 per 100000 cases of Noma [30-33]. In a study led by GESNOMA to determine the etiology of Noma in Niger, 413 cases of the disease were detected between 2001 and 2006 [32]. In addition, a six-year prospective case-control study conducted by the same group in Niger identified 82 cases of Noma to assess possible risk factors [34]. These and other studies have highlighted the burden of the disease in Niger and the so-called noma belt [33-39]. Even with the lack of recent scientific investigations on Noma, some reports confirm the occurrence of the disease in Ethiopia. For example, in three years (between 2007 and 2009), 95 patients were operated on in Addis Ababa by three visiting surgical teams from Guy's Hospital, University of Groningen. Of these, 43 patients had Noma [35, 36]. In addition, the UK-registered charity Facing Africa reported that teams of volunteer surgeons, anesthesiologists, physicians, and nurses who visit Ethiopia twice a year (usually in February and October) on two-week surgical missions have operated on 50-55 children who survived the effects of Noma since 2010 [39]. These reports suggest the disease affects many children in Ethiopia [34]. The current updated study also found a significant number of noma cases in Ethiopia. A tropical climate prevails in Afar, Somali, Gambella and Tigray regions, part of Amhara and Oromia regions where most noma cases were reported. This finding could support the argument that Noma is a neglected tropical disease. These regions accounted for nearly 83% of all patients admitted to Ethiopia's three noma treatment centers. Nevertheless, the Amhara and Oromia regions accounted for 29.2% (n=38) and 25.4% (n=33) of the noma cases registered at the three treatment facilities. The higher number of noma cases in the Amhara and Oromia regions could also be explained by the relative proximity of these regions to the three noma treatment centers and their vast population size. On the other hand, the high number of noma cases in the Amhara and Tigray regions could also be related to the largest famine in the country's history between 1984 and 1985. In addition, the fact that the region is the second most populous in the country could explain why more noma cases have settled there.

Further research needs to be conducted in this regard. Strikingly, this updated study shows that 92.9% of noma cases from all regions of Ethiopia were admitted to hospitals only at the age of 11 years and above. Inaccessibility of health care, lack of awareness, and fear of stigmatization could be primary reasons for late visits of noma cases to health facilities.

Conclusions

Understanding the national or global geographic distribution of a disease affecting a significant number of people is considered a vital impetus for addressing the problem at the national or international level. In this regard,

the results of this updated study could significantly contribute to developing a national noma prevention policy and the efficient use of available resources and manpower in Ethiopia. In addition, the scientific evidence obtained from the current study may bridge the scientific knowledge gap and serve as baseline data for further in-depth studies to better understand this mysterious disease.

Conflict of Interest

The author declares no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

References

1. Tonna JE, Lewin MR, Mensh B. A case and review of Noma. *PLoS Negl Trop Dis* 4 (2010): e869.
2. Enwonwu CO, Falkler Jr WA, Phillips RS. Noma (cancrum oris). *The Lancet* 368 (2006): 147-56.
3. Prado-Calleros HM, Castillo-Ventura BB, Jiménez-Escobar I, Ramírez-Hinojosa JP, López-Gómez A, García-de-la-Cruz M, et al. Noma and Noma-like disease in HIV/AIDS patients, a comorbid interaction: a systematic review. *J Infect Dev Ctries* 12 (2018): 89-96.
4. Berthold P. Noma: a forgotten disease. *Dent Clin* 47 (2003): 559-74.
5. Ashok N, Tarakji B, Darwish S, Rodrigues JC, Altamimi MA. A review of Noma: a current update. *Glob J Health Sci* 8 (2016): 53.
6. Adeniyi SA, Awosan KJ. Pattern of Noma (Cancrum Oris) and its risk factors in northwestern Nigeria: a hospital-based retrospective study. *Ann Afr Med* 18 (2019): 17-22.
7. Limongelli WA, Clark MS, Williams AC. Nomalike lesion in a patient with chronic lymphocytic leukemia. Review of the literature and report of a case. *Oral Surg Oral Med Oral Pathol* 41 (1976): 40-51.
8. Bello SA, Adeoye JA, Oketade I, Akadiri OA. Estimated incidence and prevalence of Noma in north-central Nigeria, 2010-2018: A retrospective study. *PLoS Negl Trop Dis* 13 (2019): e0007574.
9. Huyghe A, François P, Mombelli A, Tangomo M, Girard M, Baratti-Mayer D, et al. Microarray Analysis of Microbiota of Gingival Lesions in Noma Patients. *PLoS Negl Trop Dis* 7 (2013): e2453.
10. WHO | International Classification of Diseases (2017).
11. Marck KW. Cancrum oris and noma: some etymological and historical remarks. *Br J Plast Surg* 56 (2003): 524-7.
12. Enwonwu CO. Noma--the ulcer of extreme poverty. *N Engl J Med* 354 (2006): 221-4.

13. Giddon DB, Jerome Zackin S, Goldhaber P. Acute necrotizing ulcerative gingivitis in college students. *J Am Dent Assoc* 68 (1964): 381-6.
14. Marck KW. A history of Noma, the "face of poverty." *Plast Reconstr Surg* 111 (2003): 1702-7.
15. Gebretsadik HG, de Kiev LC. A retrospective clinical, multi-center cross-sectional study to assess the severity and sequela of Noma/Cancrum oris in Ethiopia. *PLoS Negl Trop Dis* 16 (2022): e0010372.
16. Lancet Global Burden of Disease (2017).
17. GBD 2015 DALYs and HALE Collaborators. Global, regional, and national disability-adjusted life-years (DALYs) for 315 diseases and injuries and healthy life expectancy (HALE), 1990-2015: a systematic analysis for the Global Burden of Disease Study 2015. *Lancet* 388 (2016): 1603-1658.
18. Whiteford HA, Baxter AJ. The Global Burden of Disease 2010 Study: what does it tell us about mental disorders in Latin America? *Braz J Psychiatry* 35 (2013): 111-2.
19. Gebretsadik HG. Psychosocial, Functional and Aesthetic-Related Patients' Reported Outcomes Measures (PROMS) after Orofacial Reconstructive Surgery among Noma Cases in Ethiopia. *Int J Sch Cogn Psycho* 9 (2022): 274.
20. Ja S, Cd M, S C, S S, Tb Ü, Cjl. HSPA book - healthsysperf-toc.pdf (2003).
21. Gold MR, Siegel JE, Russell LB, Weinstein. *Cost-Effectiveness in Health and Medicine - Marthe R. Gold, Joanna E. Siegel, Louise B. Russell, Milton C. Weinstein - Oxford University Press* (1996).
22. Stein C. Claudia Stein: Europe is embracing the burden of disease approach – The BMJ.
23. Bourgeois DM, Diallo B, Friehe C, Leclercq MH. Epidemiology of the occurrence of noma oro-facialis: a study of cases in Dakar, Senegal, 1981-1993. *Am J Trop Med Hyg* 61 (1999): 909-13.
24. Baratti-Mayer D, Gayet-Ageron A, Hugonnet S, François P, Pittet-Cuenod B, Huyghe A, et al. Risk factors for noma disease: a 6-year, prospective, matched case-control study in Niger. *Lancet Glob Health* 1 (2013): e87-96.
25. Oji C. Cancrum oris: its incidence and treatment in Enugu, Nigeria. *Br J Oral Maxillofac Surg* 40 (2002): 406-9.
26. Hotez PJ. *Forgotten people, forgotten diseases: The neglected tropical diseases and their impact on global health and development.* John Wiley & Sons (2021).
27. Yunusa M, Obembe A. Prevalence of psychiatric morbidity and its associated factors among patients facially disfigured by cancrum oris in Nigeria a controlled study. *Niger J Med J Natl Assoc Resid Dr Niger* 21 (2012): 277-81.
28. Farley ES, Amirtharajah M, Winters RD, Taiwo AO, Oyemakinde MJ, Fotso A, et al. Outcomes at 18 mo of 37 noma (cancrum oris) cases surgically treated at the Noma Children's Hospital, Sokoto, Nigeria. *Trans R Soc Trop Med Hyg* 114 (2020): 812-9.
29. Marck KW. Noma: a neglected enigma. *Lancet Glob Health* 1 (2013): e58-9.
30. Leila Srour M, Marck KW, Baratti-Mayer D. Noma: neglected, forgotten and a human rights issue. *Int Health* 7 (2015): 149-50.
31. Enwonwu CO. Epidemiological and biochemical studies on necrotizing ulcerative gingivitis and Noma (cancrum oris) in Nigerian children. *Arch Oral Biol* 17 (1972): 1357-71.
32. Baratti-Mayer D, Gayet-Ageron A, Cionca N, Mossi MA, Pittet D, Mombelli A. Acute necrotizing gingivitis in young children from villages with and without Noma in Niger and its association with sociodemographic factors, nutritional status and oral hygiene practices: results of a population-based survey. *BMJ Glob Health* 2 (2017): e000253.
33. Baratti-Mayer D, Baba Daou M, Gayet-Ageron A, Jeannot E, Pittet-Cuénod B. Sociodemographic characteristics of traditional healers and their knowledge of Noma: a descriptive survey in three regions of Mali. *Int J Environ Res Public Health* 16 (2019): 4587.
34. Baratti-Mayer D, Gayet-Ageron A, Hugonnet S, François P, Pittet-Cuenod B, Huyghe A, et al. Risk factors for noma disease: a 6-year, prospective, matched case-control study in Niger. *Lancet Glob Health* 1 (2013): e87-96.
35. McGurk M, Marck R. Treatment of Noma: medical missions to Ethiopia. *Br Dent J* (2010): 179-82.
36. Burki T. The fight against Noma. *Lancet Infect Dis* 16 (2016): 1231.
37. Chiandussi S, Luzzati R, Tirelli G, Di Lenarda R, Biasotto M. Cancrum oris in developed countries. *Aging Clin Exp Res* 21 (2009): 475-7.
38. Engels D. Neglected tropical diseases: An indicator of equitable development and shared prosperity. *PLoS Negl Trop Dis* 11 (2017): e0005419.
39. Rickart AJ, Rodgers W, Mizen K, Merrick G, Wilson P, Nishikawa H, et al. Facing Africa: describing Noma in Ethiopia. *Am J Trop Med Hyg* 103 (2020): 613.