

## ARTIKEL LAPORAN KASUS

### A 3-YEAR-OLD BOY FROM THE KOROWAI TRIBE WITH NOMA AND MALNUTRITION

#### NOMA DAN MALNUTRISI PADA ANAK USIA 3 TAHUN DARI SUKU KOROWAI

Tasya Gitaputri Pranoto\*, Imanuel Yulius Malino, Theo Adolf Rompas,  
I Made Agus Suarsana

Dian Harapan Hospital, Waena, Jl. Teruna Bakti, Yabansai, Kec. Heram, Kota Jayapura, Papua 99358

\* **Korespondensi:** tasya.pranoto@gmail.com

#### ABSTRACT

**Introduction:** Noma is an infectious disease that rapidly destroys soft tissues, perforating hard tissues skin of the face. Children aged 2 to 6 years with poor oral hygiene, malnutrition, malaria, HIV infection, measles, and living in poor, resource-constrained areas are at risk of the disease. This case emphasized the need for multidiscipline approaches to attain health.

**Case Report:** A 3-year-old boy from Korowai was admitted with a month's history of a lesion on the left cheek. It started as a vesicle on the left gum, progressing rapidly into an ulcer. At present, there is a hole in the left cheek. An ulcer with a diameter of 5 cm, grayish-black area, edema on the external surface of the cheek, mandibular bone, and teeth exposed. There was pain, pus, malnutrition, no immunization records, and a history of malaria.

The patient was managed by blood transfusion, antibiotics, debridement of the affected area, and a high carbohydrate and protein diet. After the lesion was more minor, no other complications were revealed, and the patient was discharged.

**Conclusion:** Noma is an abandoned disease known as the "face of poverty." Early intervention can be made to minimize tissue destruction. Adequate steps regarding the availability, accessibility and utilization of the resources with a cultural approach might help to improve the health conditions of individuals living in susceptible areas.

**Key Words:** infection, Korowai, malnutrition, noma, tropical disease

#### ABSTRAK

**Pendahuluan:** Noma adalah penyakit infeksi langka yang menyerang jaringan lunak, hingga perforasi tulang dan kulit wajah. Anak usia 2 hingga 6 tahun dengan sanitasi rendah, malnutrisi, malaria, HIV, campak, tinggal dalam kemiskinan, lingkungan yang jauh dari fasilitas merupakan faktor risiko dari noma. Kasus ini menunjukkan bahwa diperlukan pengertian, pendekatan multidisiplin untuk mencapai status sehat.

**Laporan Kasus:** Anak laki-laki, usia 3 tahun dari Korowai datang dengan keluhan lesi di pipi kiri sejak 1 bulan. Berawal dari vesikel di gusi kiri yang berkembang menjadi ulkus. Saat pemeriksaan ditemukan lubang pada pipi kiri pasien. Sebuah ulkus diameter 5 cm disertai area abu kehitaman, edema permukaan luarnya, tulang mandibula, gigi pasien dari luar. Keluhan disertai nyeri dan nanah di tempat luka. Pasien datang dengan malnutrisi, tidak ada status imunisasi, riwayat sering terkena malaria sebelumnya.

Tatalaksana meliputi transfusi darah, antibiotik, debridemen pada daerah yang terinfeksi, dan diet tinggi protein dan karbohidrat yang sesuai dengan makanan lokalnya seperti pisang, telur, dan sagu. Pasien dipulangkan setelah lesi membaik dan komplikasi teratasi.

**Simpulan:** Noma adalah salah satu penyakit yang lama ditinggalkan, disebut sebagai "Face of Poverty". Intervensi dini dapat dilakukan untuk mengurangi kerusakan jaringan. Diperlukan tatalaksana meliputi ketersediaan, aksesibilitas, utilisasi sumber daya, dengan pendekatan budaya untuk membantu meningkatkan kesehatan masyarakat yang tinggal di area yang bersangkutan.

**Kata Kunci:** infeksi, Korowai, malnutrisi, noma, penyakit tropis

#### INTRODUCTION

Noma is a rare infectious disease that rapidly destroys the soft tissues, perforating

the face's hard tissues and skin. It was first named in the eighteenth century. The world prevalence of noma is unknown, and

estimates range from 30.000 to 140.000 cases. Most cases were found across Africa, from Senegal to Ethiopia.<sup>1</sup> Children aged between 2 and 6 years with poor oral hygiene, malnutrition, malaria, HIV infection, measles, living in poor conditions, and resource-constrained areas are at risk of the disease. The pathogenesis of noma is unknown, but studies show interactions of several factors, including infectious agents (bacterial and viral), malnutrition, and immune suppression, are essential for its development. Noma is not contagious and not recurrent. If left untreated, the condition may be fatal, and patients suffer facial destruction that requires major surgical reconstruction.<sup>2</sup>

Malnutrition is one of the common problems afflicting low- and middle-income countries. This problem is one of the utmost concerns in this Sustainable Development Goals (SDGs) era. Malnutrition, poverty, and chronic diseases are interconnected so that each of the factors affects the other, resulting in an undeniable impact.<sup>3</sup> In Indonesia, malnutrition is associated with stunting, being underweight, and being overweight. Seven provinces have a very high prevalence (>40%), and 17 provinces have a high prevalence (30-39%) of stunting. Tenggara, Papua, and West Java have been the most vulnerable to poverty and food insecurity.<sup>4,5</sup>

Korowai are the people who live in southeastern West Papua, Indonesia. Close to the border of Papua Guinea. The majority of the clans live in the treehouses on their isolated territory. They live by hunting and fishing. Over the years, these clans had lived

separately within a few kilometers of each family. They only gather to do rituals together.<sup>6</sup> This case emphasizes the need for awareness and greater understanding not only of the disease but also the diverse approach to attain health.

## CASE REPORT

A 3-year-old boy from Korowai was admitted with a month's history of a lesion on the left cheek. The lesion was started as a vesicle on the left gum, progressing rapidly into an ulcer. At present, there is a hole in the left cheek. The lesion was started as an ulcer with a diameter of 5 cm, a grayish-black area, and edema appeared on the external surface of the cheek, mandibular bone, and teeth exposed. Associated with the lesion were pain, pus, no immunization records, and a history of recent malaria. Other complaints were coughing and sneezing 1 week before the admission. The family came barefooted. Other past histories were not able to be collected because of language boundaries. The family only spoke the local language. The patient lived with his parents, a brother, and a sister in a treehouse at Korowai. Hunting was a way of their living and surviving.

Physical examination revealed heart rate was 108x/minute, respiratory rate 26x/minute, and the temperature was 37.5°C. Pale conjunctiva and poor oral hygiene were reckoned, and rhonchi were found at the basal of the lungs. No significant findings on the cor and abdomen evaluation. His height was 89 cm, weighed 10 kg. Height for age was -1.76 SD, weight for age -2.65 SD, and weight for

height was -3 SD. Laboratory findings were hemoglobin was 5,5 g/dL, microcytic hypochromic anemia, elevated erythrocyte sedimentation rate, and normal leukocytes. Radiology findings were infiltration of the lungs and

no mass or infiltration of the mandibula or facial bone on the skull X-ray. Blood and pus culture was taken. The patient was admitted to the pediatric ward. The head and neck surgeon was consulted on the next day.



**Figure 1.** Noma before operation:  
(a) A hole on the left cheek when first admitted;  
(b) The lesion on the 7<sup>th</sup> day of admission

The patient was given maintenance fluid, Ceftriaxone 500 mg/12h, Gentamicin 2x20 mg, Sodium metamizole 250 mg/8h, and a blood transfusion. He was put on a high carbohydrate and protein diet with 1200 kcal per day during the hospitalization. On the first two days of admission, the patient would not eat anything served. No rice or milk, not even meat. Later we observed that he would only eat the eggs. Then diet was changed to bananas, eggs, and sago, his local foods. The wound was dressed twice per day with honey and measured every day. The surgery was rescheduled.

On the third and fourth days of admission, cough and rhonchi were still found at the base of the lungs. Multivitamins syrup, ibratropium bromide and salbutamol inhalation/8h, paracetamol injection was given. The Mantoux test

was negative, and no HIV infection was found. The tuberculosis scoring of pediatricians was 4. On the fifth day of admission, rhonki was decreased, and fever was documented with the highest temperature of 39°C. Elevated white blood count ( $13,65 \times 10^3/\text{ml}$ ) and hypoalbuminemia (2,4 g/dL) were found. Albumin transfusion was given. On the seventh day of admission, the lesion got smaller than before, sized 2x3cm. Peripheral edema around the lesion was fading.

On the eleventh day of admission, fever was still found. No nausea, no vomit, and no headache were found. Daily intake was still good. The patient was then diagnosed with malaria. Plasmodium falciparum was found under the microscope. Artesunate 24 mg injection was given three times (0-12-24) and Primaquin 1 x 1/4 tablet.

After the lesion got smaller, infection was controlled, albumin was improving, no edema surrounding the lesion area, no fever was documented, and surgery was conducted on the fourteenth day of admission. The procedure was debridement of the affected

area, including two teeth extraction on the infected area. The postoperative wound was dry, no pus, no edema was found, and no other complication was revealed after the surgery. On the 21<sup>st</sup> day, the patient was discharged.



**Figure 2.** Noma after surgery:

- (a) Postoperative wound
- (b) The local wound had essentially healed after discharge

## DISCUSSION

This report aims to expose a noma case found in Papua, Indonesia. A disease that was unheard of and under-detected globally, resulting in a lack of experience, information, and awareness about one. This patient was brought from Korowai, a tribe in Asmat District, by a missionary to the local health facility, then referred to the secondary hospital after a month since the first ulcer appeared. The causative agent of noma remains unknown. Reksodiputra, in his literature review, mentioned that there are several species of bacteria often found with noma due to rapid evolution and the odor from the lesions, which are *Borrelia vincentii* and *Fusobacterium fusiformis*.<sup>7,8</sup> In this case, blood and pus cultures could not be done due to a lack of

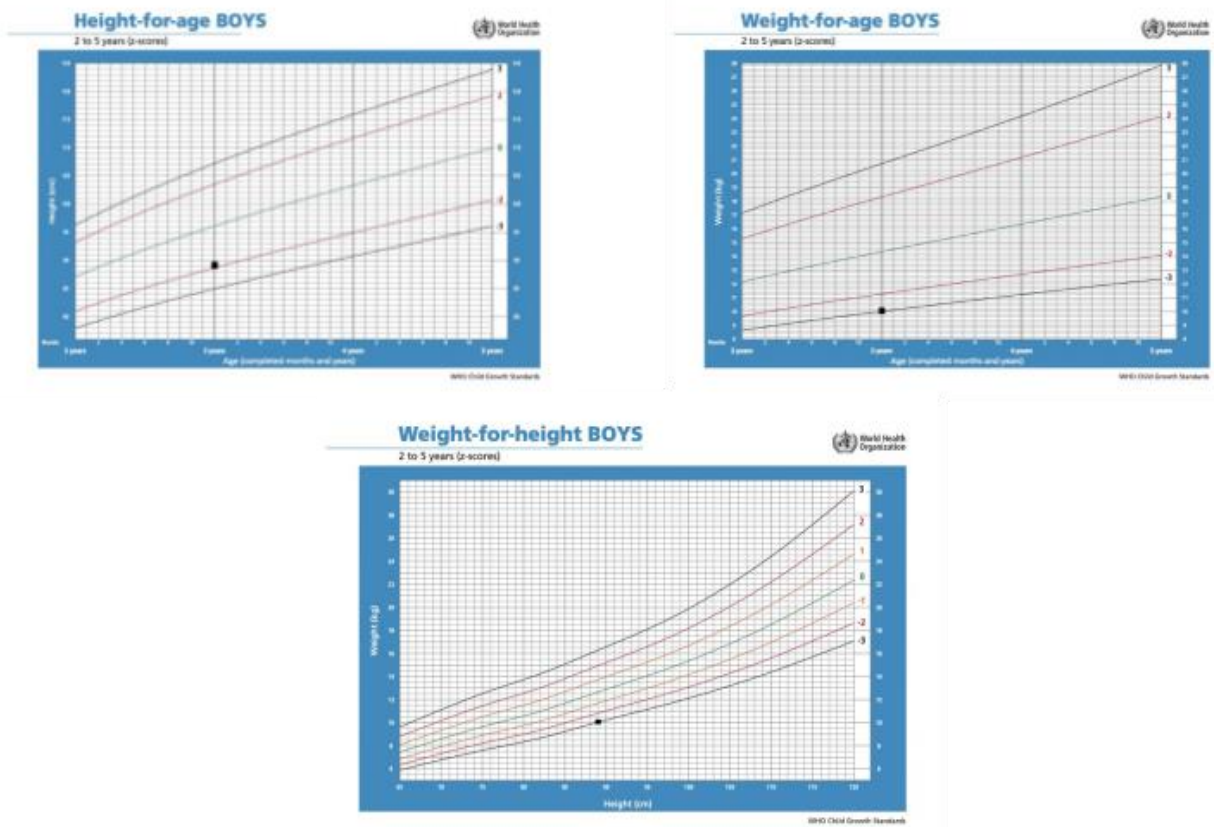
facility and resources. Other studies suggest the etiology of noma as an opportunistic multifactorial that occurs in the presence of normal oral flora in chronically malnourished children.<sup>8,9</sup>

Malnutrition is defined as an imbalance between nutrient requirement and intake, resulting in cumulative deficits of energy, protein, or micronutrients that may negatively affect growth, development, and other relevant outcomes, according to the American Society of Parenteral and Enteral Nutrition (ASPEN). Malnutrition is either illness-related (diseases or injuries directly result in nutrient imbalance) or caused by environmental or behavioral factors associated with decreased nutrient intake and delivery. Most noma cases were found associated with malnutrition.<sup>3,8,9</sup>

According to the WHO growth chart for this patient, the weight for height chart Z score was at -3 SD, which implies severe acute malnutrition.

Height for age is one of the indicators of nutritional problems. It shows chronic nutritional problems resulting from circumstances in the long term, for example, poverty, un-

healthy lifestyle, and less feeding since newborn that cause children to be short. The other indicator is also Weight for Height, which indicates the nature of acute nutritional problems. Among 33 provinces in Indonesia, 19 provinces had a prevalence of underweight children above the national prevalence rate, ranging from 19.7% to 33.1%.<sup>4,10,11</sup>



**Figure 3.** WHO Growth Chart for this patient

Anemia, which reflects several micro-nutrient deficiencies, was found in this patient. Microcytic hypochromic depicts a lack of iron intake in the body. The other finding was hypoalbuminemia which also implies a lack of protein problem. Like other low- and middle-income countries, despite Indonesia's sustained economic growth and poverty reduction, malnutrition is still far from being

over. Unavailability, little or even no access to food, lack of dietary diversity, social beliefs and traditions, low education, and high poverty have been the diverse cause of malnutrition.<sup>9,10,13</sup> In this case, the patient only ate local foods such as sago, banana, and eggs. Other than that, the food that was given remains untouched. This response depicts a lack of disparities in the diet. Assuming it may

lead to a lack of carbohydrates, proteins, vitamins, and minerals needed for the body, which might happen for a long time. Chronically malnourished children are at risk for opportunistic infections by the weakening of the immune system and increased

susceptibility of the body to infections. Malnourished children are more prone to die of respiratory infections and diarrhea, thus increasing mortality and morbidity rate. Not only infections but also negatively influence human cognitive and productivity.<sup>3,9</sup>

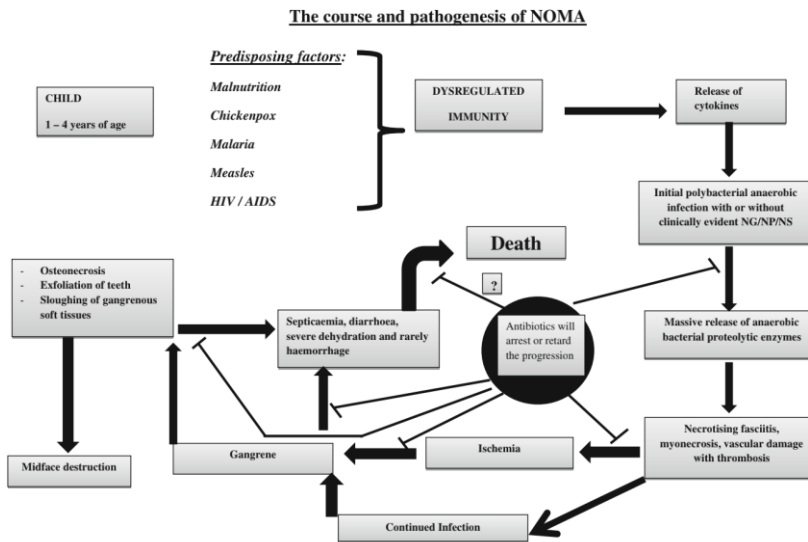


Figure 6. The pathogenic events in noma<sup>2</sup>

Unlike this case, a case found in India, a 2-year-old boy presented with recurrent fever, lymphadenopathy, thrombocytopenia, leukopenia, appropriate immunization, and asymptomatic lesion of the right cheek with discharging sinus.<sup>11</sup> In this current case, a 3-year-old boy with a lesion on his left cheek with pain and no fever. The majority of the Korowai tribe, where this patient is from, drink from the rain and river without cooking it first. They do not have in-house toilets, defecation is carried out in the forests and rivers near their houses, as well as bathing and cleaning food.<sup>12</sup>

According to Figure 6 which shows the pathogenesis of noma, assuming malnutrition, recent malaria, and poor oral hygiene as the cause of dysregulated immunity which

possibly initiates polybacterial anaerobic infection with necrotizing gingivitis.<sup>(2,11)</sup> Untreated, it extends to become necrotizing periodontitis and then necrotizing stomatitis. Necrotizing stomatitis is characterized by the destruction of oral soft tissues and bone. Without treatment, the necrotizing process will spread rapidly to the mucosa of the cheek or lip, muscle and skin. A massive release of anaerobic bacterial proteolytic enzymes causes necrotizing fasciitis, myonecrosis, and vascular damage. The magnitude of tissue damage depends on the fitness and virulence of the microbial community, the general health of the host, and the fitness of the host's immune system.<sup>8,11,13,14</sup> Secondary immunodeficiency due to HIV was ruled out.

Any treatment of any stage in the sequence of necrotizing gingivitis, necrotizing periodontitis, and necrotizing stomatitis will prevent the development of noma. Treatment includes appropriate orally administered antibiotics, frequent use of chlorhexidine mouthwash, toothbrushing, and analgesic. Once the acute phase has been controlled, debridement of roots, necrotic soft tissue, and bone, and removal of hopelessly mobile teeth should follow. Once the necrotizing process has progressed to noma, a treatment protocol similar to that should be introduced immediately, only after the general condition of the patient has been stabilized, tissue debridement and tooth extraction should be performed.<sup>13-15</sup> Fortunately the process did not go further to septicaemia in this case. The patient was given broad spectrum antibiotics for infection, packed red cells for anemia, and nutritional supplementation as early as he was admitted to the hospital. The wound was dressed with honey twice daily. Yilmaz review reported that honey in acute and chronic wounds provided rapid epithelialization and wound contraction in wound healing, had anti-inflammatory and debridement effects, decreased the pain, ensured infection control, shortened the time of wound healing. Another advantage honey was cost-effective.<sup>15</sup>

Children from low-income households and those who have less educated parents likewise are at a higher risk for negative health outcomes and disease development. Lack of sanitation also plays a role in attaining health. In addition to that, most of them have limited access to healthcare facilities which end up in

delay or neglect to seek medical treatment.<sup>6,8,11</sup> Noma can be avoided through simple actions that can be performed by anyone. Opening and examining our children's mouths, and regular oral examination of children at home or during medical visits is an action that helps to identify gum lesions that may develop into noma in at-risk subjects. It is important to understand that noma is not caused by witchcraft or curse on the family.<sup>11,13,16</sup>

## CONCLUSION

NOMA is an abandoned disease known as the "face of poverty." Early intervention can be made to minimize tissue destruction. Nevertheless, adequate steps regarding the availability, accessibility and utilization of the resources with a cultural approach might help to improve the health condition of individuals living in susceptible areas.

## REFERENCES

1. Marck KW. Noma: a neglected enigma. *Lancet Glob Health*. 2013;1(2):e58-e59
2. Masipa JN, Baloyi AM, Khammissa RA, Altini M, Lemmer J, Feller L. Noma (cancrum oris): A report of a case in a young AIDS patient with a review of the pathogenesis. *Head Neck Pathol*. 2013;7(2): 188-192
3. Adebisi, Y.A. *et al*. Prevalence and socio-economic impacts of malnutrition among children in Uganda. *Nutrition and Metabolic Insights*. 2019
4. Tosepu R, Effendy DS, Bahar H, Sakka A, Asfian P, Lestari H. Did Indonesia achieve the MDGs goals by 2015? *Public Health of Indonesia*. 2016;30;2(1):1-9
5. Schröders J, Wall S, Kusnanto H, Ng N. Millennium development goal four and child health inequities in Indonesia: A systematic review of the literature. *PLoS ONE*.2015;10(5):e0123629

6. Reksodiputro M.H, Yosia M. Noma: a neglected tropical disease. *ORLI*. 2021;51(1):56-63
7. Ashok, N., Tarakji, B., Darwish, S., Rodrigues, J.C., Altamimi, M.A. A review on noma: A recent update. *Global journal of health science*. 2015;8(4), 53–59
8. Baratti-Mayer D, et al. Geneva Study Group on Noma (GESNOMA). Risk factors for noma disease: a 6-year, prospective, matched case-control study in Niger. *Lancet Glob Health*. 2013;1(2):e87-e96.
9. Dipasquale V, Cucinotta U, Romano C. Acute malnutrition in children: pathophysiology, clinical effects and treatment. *Nutrients*. 2020;12(8):2413
10. Taki, Sarah. Malnutrition among children in Indonesia: It is still a problem. *JKKI [Online]*:2018:68-71.
11. Feller L, Khammissa RA, Altini M, Lemmer J. Noma (cancrum oris): An unresolved global challenge. *Periodontology 2000*. 2019;80(1):189-99.
12. Singh A, Mandal A, Seth R, Kabra SK. Noma in a child with acute leukaemia: When the 'face of poverty' finds an ally. *BMJ Case Rep*. 2016;bcr2015211674
13. Kaimudin, A., & Hidajah, A. Epidemiological investigation of noma in Papua province. *Jurnal Berkala Epidemiologi*, 2017;8(1), 16-25
14. Huyghe A, François P, Mombelli A, Tangomo M, Girard M, Baratti-Mayer D, Bolivar I, Pittet D, Schrenzel J, Geneva Study Group on Noma. Microarray analysis of microbiota of gingival lesions in noma patients. *PLoS Negl Trop Dis*. 2013;7(9):e2453
15. Xu, L., Wei, W., Ge, X. et al. Noma in a boy with septic shock: a case report. *BMC Pediatr*. 2019;19:200
16. Yilmaz AC, Aygin D. Honey dressing in wound treatment: A systematic review. *Complement Ther Med*. 2020;51:102388.
17. World Health Organization. Regional Office For Africa [Internet]. Noma is a severe disease. It is treatable if detected and managed early. 2016. Available from [https://www.afro.who.int/sites/default/files/2017-07/Information\\_brochure\\_EN.pdf](https://www.afro.who.int/sites/default/files/2017-07/Information_brochure_EN.pdf)