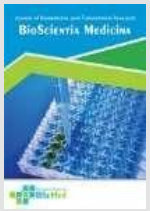
eISSN (Online): 2598-0580



Bioscientia Medicina: Journal of Biomedicine & Translational Research

Journal Homepage: www.bioscmed.com

Difficult Airway Management of Reconstructive Surgery for Noma (Cancrum oris): A Rare Neglected Disease

Benny Supono[1*], Adinda Putra Pradhana[1], I Gusti Putu Sukrana Sidemen[1], Putu Kurniyanta[1]

[1]Department of Anesthesiology and Intensive Care, Faculty of Medicine, Universitas Udayana/Prof. Dr. I.G.N.G. Ngoerah General Hospital, Denpasar, Indonesia

ARTICLE INFO

Keywords:
Cancrum oris
Difficult airway
Fiberoptic intubation
Nasal fiberoptic intubation
Noma

***Corresponding author:**
Benny Supono

E-mail address:
Bennysupono@student.unud.ac.id

All authors have reviewed and approved the final version of the manuscript.

https://doi.org/10.37275/bsm.v8i3.937

A B S T R A C T

Background: Noma is a rare necrotizing gangrenous stomatitis that occurs due to poor oral hygiene and chronic malnutrition. Noma's survivors usually had significant facial deformities that needed reconstructive surgery as its definitive treatment. However, this facial deformity can result in a difficult airway that is very challenging for anesthesiologists. **Case presentation:** A 22-year-old male patient had a significant deformity on his left face due to Noma. Preoperative evaluation revealed a potentially difficult airway due to deformity of the maxilla and mandible, malocclusion, inadequate mask seal, and incomplete dentition. Nasal fiberoptic intubation was chosen as the management of a difficult airway in this patient. A tracheostomy was prepared as the emergency invasive airway in the event of failed intubation attempts. Intubation attempts were limited to three times, and the nasal fiberoptic intubation in this patient was successful on the third attempt. The patient was stable, and the airway was safely maintained during the surgery. **Conclusion:** Detailed and careful perioperative evaluation had vital role assessing potential difficult airway and planning the optimal airway management for patient with facial deformity. Nasal fiberoptic intubation is still the safest choice with high success rate for Noma patient with significant facial deformity.

1. Introduction

Noma (Cancrum oris) is a rare necrotizing orofacial gangrene that could rapidly destroy other facial structures. Noma is usually found in children with chronic malnutrition, poor oral hygiene, and lack of healthcare access; thus, this disease is often considered "the face of poverty". Several decades earlier, Noma could be found worldwide, including the developed countries such as America and Europe. However, along with increasing welfare and advanced health technology, Noma become a very rare disease that is only found in countries with extreme poverty and chronic malnutrition. Today, Noma is mostly found in African and Sub-Saharan children, and literature regarding Noma cases in Indonesia is very limited.[1–3]

The exact pathogenesis of Noma is unknown, but progressive polymicrobial opportunistic infections are considered as the main cause of Noma. Noma had a high mortality rate (80-90%) during the acute phase, with most of the patients dying due to severe malnutrition, dehydration, and sepsis during the acute phase.[3,4] Survivors of this acute phase will live with significant maxillofacial deformities that need reconstructive surgery.[5,6] Airway management in patients with abnormal maxillofacial structures is very challenging for an anesthesiologist. Most Noma patients have difficulty opening the mouth while the

surgery process requires extensive orofacial manipulation, so perioperative airway management strategies must be carefully planned. Preoperative evaluation plays an important role in predicting a potential difficult airway during maxillofacial manipulation in patients with Noma.[5,7] Therefore, we report a successful nasotracheal intubation using fiberoptic laryngoscope to maintain the airway during the reconstructive surgery in an adult patient with Noma.

2. Case Presentation

A 22-year-old male patient presented with significant facial deformity as the sequalae of Noma (Cancrum oris). Patient had difficulty of mouth opening but he also cannot close his lower jaw properly since he was 2-year-old. Patient also said that the corner of his left lip was pulled upwards since he was 3-years-old. These complaints began with a small lump at the size of a green bean on the patient's left lip area when he was 2-years-old. The lump became enlarged within 1.5 months and then a whitish smelly fluid comes out from the lump after it ruptured. A wound with gangrenous slough was formed and it became wider and deeper until it reaches the gums. The patient received traditional herbal ointment for 7 months. The wound was resolved, but his lips was pulled upwards and he had difficulty and pain when moving his jaw. during preoperative evaluation, patient said that he had limited jaw movement due to the deformity in the maxillofacial structure. No history of systemic disease, allergic, or prior surgery in this patient.

The patient weighs 49 kg, height 167 cm, and a body mass index (BMI) of 17.7 kg/m^2. The vital signs and general physical examination were within normal limits, but the orofacial examination revealed a deformity in the left maxilla and mandible region accompanied by trismus. Airway evaluation revealed that the interincisor distance was less than 1 finger, mentohyoid distance was 3 finger breadths, thyrohyoid distance was 2 finger breadths (Figure 1). There were also mandibular malocclusion and incomplete dentition. Thus, the LEMON score was 7/10. Assessment of MOANS score showed an inadequate mask seal. However, Mallampati score was difficult to evaluate. Therefore, we conclude there was potential difficult airway in this patient.

Routine laboratory tests were within normal limit. Plain cervical X-rays in AP/Lateral view showed a deformity in left mandibular body with thickening of the cortex due to chronic inflammation. Based on computed tomography (CT)-scan of the head, there was hyperdense cutis-subcutis of left maxilla until buccal area which was suspected as cicatrix. There was also deformity in the left maxillary alveolar process and left mandibular body deformity of left maxillary alveolar process, but no ankylosis or fusion found on left and right temporomandibular joint with a conclusion of type II malocclusion (overbite). To anticipate the potentially difficult airway with limited jaw movement, we planned to use nasal fiberoptic intubation as the first line and emergency tracheostomy as the second line if the intubation attempts failed.

Figure 1. Preoperative airway evaluation.

As premedication, oxymetazoline nasal drop was instilled in the right nose and patient was nebulized with 4% lidocaine for 10 minutes. The patient was transferred to the operating room, and standard monitoring equipment (including non-invasive blood pressure, heart rate, respiratory rate, and peripheral pulse oximeter) was installed. Before induction, patient was given 150 mcg fentanyl and 80 mg lidocaine IV. Induction was performed using 100 mg Propofol and continuous dexmedetomidine with a maintenance dose of 0.2-0.7 mcg/kgBW/hour. When the patient has begun to be hypnotized but not apneic, a flexible intubating scope (FIS) was inserted through right nostril. When epiglottis, glottis, and vocal cord had been visualized, FIS was inserted further into the trachea until the carina was visualized through FIS. A size 7.0 non-kinking endotracheal tube (ETT) was inserted using FIS guidance to a depth of 24 cm at the level of right nostril. After the ETT position was confirmed to be symmetrical, patient was given 30 mg rocuronium IV. Anesthesia was maintained using oxygen, continuous Propofol drip with a dose of 50-150 mcg/kgBW/minute, and intermittent fentanyl with a dose of 0,25 mcg/kgBW every 30-60 minute IV. Patient also received 1000 mg paracetamol IV and 400 mg Ibuprofen IV as analgesia. Duration of surgery was 4 hour and 40 minutes. Patient was hemodynamically stable during the perioperative period. Systolic blood pressure range was within 100-140 mmHg and diastolic blood pressure range was within 50-80 mmHg. Heart rate fluctuation during surgery was within 65-88 beats/minute and SpO_2 ranged between 97%-99%. During surgery, total blood loss was 300 ml, urine output was 700 ml, and total crystalloid fluid given was 1200 ml.

Extubation was delayed and patient was transferred to the intensive care unit (ICU) postoperatively. Patient using PC-BIPAP ventilator with FiO_2 40%, inspiratory pressure (Pins) 15, respiratory rate 12 times/minute, positive end-expiratory pressure (PEEP) 5, pressure support (PSupp) 8, and SpO_2 100%. Postoperative analgesia regimen of this patients includes 300 mcg fentanyl in 50 ml NaCl 0.9% that given with a rate of 2.1 ml/hour IV and 1000 mg paracetamol every 8 hours IV. Weaning was done gradually and patient was successfully extubated at 20 hours postoperative. Patient was then transferred to intermediate ward on the second day postoperative and discharged on fifth days postoperative. No postoperative complications or adverse effects found in this patient.

3. Discussion

Difficult airways area always challenging a major problem in surgery involving the oral and maxillofacial areas, whether caused by trauma, infection, or tumors and malignancies in the maxillofacial area. American Society of Anesthesiologists (ASA) the clinical situation in which anticipated or unanticipated difficulty or failure is experienced or trained anesthesiologist. Those clinical situations including, but not limited to, one or more of the following: face-mask ventilation, laryngoscopy, ventilation using a supraglottic airway, tracheal intubation, extubation, or invasive airway. Management of difficult airway begins with detailed and careful airway preoperative airway evaluation to anticipate and developed strategies that will be used to maintain the airway during perioperative period. Several things that must be explored in the preoperative history to estimate airway difficulties are age, use of dentures or absence of teeth, history of previous difficult intubation, history of snoring, and comorbidities (diabetes mellitus, heart disease, etc.). Preoperative physical examination should include BMI, measurement of facial and jaw parameters (including ability to open the mouth, mobility of the neck and head, evaluation of the presence of prominent upper incisors, presence of beard, and upper lip bite test), anatomical measurements of the jaw and neck (including evaluation of Mallampati score, LEMON score, MOANS score). Additional evaluation using radiological examination (head CT scan, bedside endoscopy, virtual laryngoscopy/bronchoscopy) may help to characterize the difficult airway encountered.[8,9]

Our patient had maxillofacial deformity due to extensive infected wounds in the oral area, resulting in difficulty opening and closing the jaw. Preoperative evaluation showed that this patient was hemodynamically stable with no systemic diseases or complications. However, airway evaluation showed a potential difficult airway where the Mallampati score was difficult to evaluate, there was mandibular malocclusion, LEMON score 7/10, inadequate mask seal, and incomplete dentition. The potential for a difficult airway was also proven by the results of a head CT scan which showed left-sided facial deformity with the conclusion of type II malocclusion (overbite).

The next step after airway evaluation is to choose the strategy that will be used to manage the difficult airway. To date, there are several algorithm guidelines for managing difficult airways. The ASA difficult airway guidelines were the first difficult airway management guidelines recognized internationally in 1993. These guidelines are continuously updated and are the most widely used guidelines throughout the world. In general, if difficult laryngoscopy is suspected during preoperative evaluation, but the patient had low risk of aspiration, low risk of rapid desaturation, and no difficulty in expected in performing invasive emergency airway (such as surgical tracheostomy or cricothyrotomy), then an intubation attempt can be made using either the awake intubation technique or intubation after general anesthesia induction. However, if laryngoscopy, intubation and invasive emergency airway are suspected to be difficult or the patient had high risk of aspiration and high risk of rapid desaturation, thus awake intubation is recommended. There are several alternative intubation techniques offered in the ASA difficult airway management algorithm, such as awake fiberoptic intubation, intubation with induction using general anesthesia, awake oral laryngoscopy, or videolaryngoscope. If the surgical procedure is performed in the maxillofacial area, intubation could be performed using other approaches, for example nasotracheal or submental intubation approaches. According to the algorithm shown in Figure 1, if the first intubation attempt fails, consider limiting the number of intubation attempts while seeking help or preparing for emergency airway invasion.[8,10,11]

To anticipate a difficult airway in this patient, intubation was planned using nasal fiberoptic intubation. The nasal route was chosen because the patient had difficulty opening the jaw so it was estimated that laryngoscopy would be difficult. Even though nasal fiberoptic intubation was planned from the start, anticipation of an emergency airway in the form of a tracheostomy was also prepared in the operating room to anticipate intubation failure. We consider tracheostomy as the second option because this operation is a single stage surgery.

Previous literatures also used nasal fiberoptic intubation to anticipate difficult airway in patients with facial deformity due to Noma. Braun et al. (2020) demonstrated the successful use of the nasal fiberoptic intubation technique in all 65 patients with Noma in Africa. The fiberoptic intubation was the safest option for Noma which have high success rate (97.1-99.6%) in various difficult airway cases. The use of oral route was not recommended in patient with Noma thus conventional intubation techniques that require oral approach (such as videolaryngoscope and use of a supraglottic airway) should not be used in patient with Noma.[5] The efficacy of the nasal fiberoptic intubation technique was also proven in the study of Coupe et al. (2013) where this technique was successfully performed in 8 patients and adequate ventilation was achieved in all patient. Videolaryngoscope and direct laryngoscopy technique were also successfully use in Noma patients with minimal oral and maxillofacial deformity in the study of Coupe et al. However, there were two patients with failed intubation when using video-laryngoscope technique thus the intubation technique was changed to the fiberoptic intubation technique.[6] Deo et al. (2016) also succeeded in maintaining the airway in Noma patients with limited jaw movement using a retrograde intubation technique with a central venous catheter.

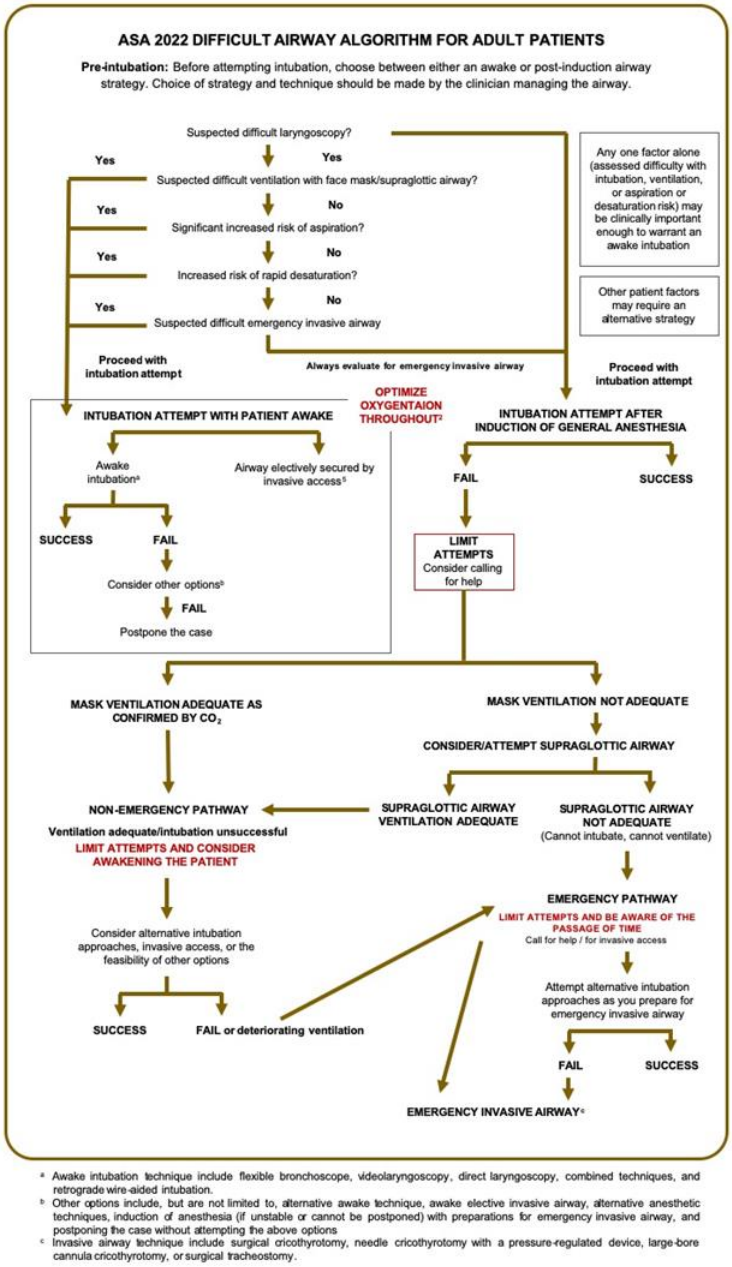


Figure 2. ASA 2022 difficult airway management algorithm for adult patient.[8]

This retrograde intubation technique uses the Seldinger technique to reach the cricothyroid membrane to perform intubation successfully. This retrograde intubation technique has several risks of complications, such as laryngeal trauma due to the wire or needle used, bleeding, hematoma, esophageal puncture, subcutaneous emphysema, infection, and pneumomediastinum. Contraindications to this technique include impossibility of cricothyroid area abnormalities, significant coagulopathy, and infection of the cricothyroid area. Fiberoptic intubation technique was not used in Deo et al. due to limited resources and high cost. The blind nasal intubation technique is not preferred because it has a high failure rate and repeated attempts will increase the risk of airway trauma and edema which can result in airway obstruction.[7]

Although the nasal fiberoptic intubation failed on the first and second attempt, intubation was successfully performed at the third attempt in our

patient. We limited intubation attempts to three times and we also have prepared emergency tracheostomy as the emergency invasive airway which is consistent with ASA algorithm for difficult airway in adult patient (Figure 2). However, it was proven nasal fiberoptic intubation capable to overcome and maintain the airway in our patient with no postoperative complications. The patient was safely extubated 20 hours postoperatively and patient was discharged 5 days postoperatively.

4. Conclusion

Difficult airway is the most common problem encountered in cases with maxillofacial trauma or maxillofacial deformity. Difficult airway is also commonly found in Noma patients due to significant maxillofacial tissue defects. A detailed preoperative evaluation and careful preparation will help the anesthesiologist in anticipating and preparing for the worst possibility (including preparing for an invasive emergency airway if necessary). To date, it has been proven that the nasal fiberoptic intubation technique is still the best option with the best level of safety and efficacy for airway management in Noma cases.

5. References

1. Farley E, Mehta U, Leila Srour M, Lenglet A. Noma (Cancrum oris): a scoping literature review of a neglected disease (1843 to 2021). PLoS Negl Trop Dis. 2021; 15(12): 1–23.
2. Brahmantara BN, Bagus I, Kartha M, Budiana ING. Characteristic gynaecology-oncology patients from obstetrics and gynaecology outpatient clinic Mangusada Regional Hospital Badung reffered to Sanglah Hospital from 1st January 2016 – 31st December 2018. Bali Med J. 2020; 9(2): 430–6.
3. Feller L, Khammissa RAG, Altini M, Lemmer J. Noma (Cancrum oris): an unresolved global challenge. Periodontol 2000. 2019; 80(1): 189–99.
4. Rajaleelan W, Otterman M. Airway Management in children with noma sequelae undergoing craniomaxillofacial reconstructive surgery. J Neuroanaesth Crit Care. 2020; 07(21).
5. Braun U, Wiese KG, Merten HA, Timmermann A. Anaesthetic care for noma (Cancrum oris) - the disease, the airway and how to provide anaesthetic care without a clinical safety infrastructure. Trends Anaesth Crit Care. 2020; 31: 16–20.
6. Coupe MH, Johnson D, Seigne P, Hamlin B. Airway management in reconstructive surgery for noma (Cancrum oris). Anesth Analg. 2013; 117(1): 211–8.
7. Deo GP, Shrestha SK, Neupane S, Ojha H, Shrestha IN, Regmi B, et al. Retrograde intubation in a case of cancrum oris with sygnathia. J Chitwan Med Coll. 2016; 6(17): 62–6.
8. Apfelbaum JL, Hagberg CA, Connis RT, Abdelmalak BB, Agarkar M, Dutton RP, et al. 2022 American Society of Anesthesiologists Practice Guidelines for management of the difficult airway. Anesthesiology. 2022; 136: 31–81.
9. Edelman DA, Perkins EJ, Brewster DJ. Difficult airway management algorithms: a directed review. Anaesthesia. 2019; 74(9): 1175–85.
10. Costan VV, Ciofu ML, Comanescu MP, Sava FP, Patrascanu E, Hartie VL, et al. Management of difficult airway in oral and maxillo-facial surgery. Rom J Med Dent Educ. 2020; 9(5): 46–53.
11. Karim HMR, Panda CK, Arora P, Basumatary K. Fibreoptic intubation under conscious sevoflurane sedation in anticipated difficult intubation cases with unfavorable conventional airway preparation. Bali J Anesthesiol. 2019; 3(2): 137–9.