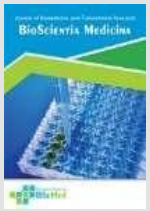
eISSN (Online): 2598-0580



Bioscientia Medicina: Journal of Biomedicine & Translational Research

Journal Homepage: www.bioscmed.com

Successful Anesthetic Management for Mandibular Neoplasm Resection in a Patient with Osteogenesis Imperfecta: A Case Report

Andi Riza Mirda Indriani[1*], T Addi Saputra[1], Novita Anggraeni[1]

[1]Department of Anesthesiology and Intensive Therapy, Faculty of Medicine, Universitas Riau/Arifin Achmad Regional General Hospital, Pekanbaru, Indonesia

ARTICLE INFO

Keywords:
Anesthesia
Case report
Difficult airway
Mandibular neoplasm
Osteogenesis imperfecta

***Corresponding author:**
Andi Riza Mirda Indriani

E-mail address:
andiriza1995@gmail.com

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

https://doi.org/10.37275/bsm.v8i12.1140

A B S T R A C T

Background: Osteogenesis imperfecta (OI) is a rare genetic disorder characterized by bone fragility and deformities. Patients with mandibular neoplasms and suspected OI require careful anesthetic management to avoid complications. This case report describes the successful anesthetic management of a patient with a mandibular neoplasm and suspected OI. **Case presentation:** A 33-year-old man presented with a large mandibular tumor causing airway obstruction and difficulty eating. He had physical features suggestive of OI, including short stature, bone deformities, and abnormal tooth growth. Preoperative evaluation revealed a difficult airway due to the tumor and potential cervical spine instability. Anesthesia was induced with propofol and atracurium after securing the airway via ultrasound-guided tracheostomy under local anesthesia. The tumor was resected successfully, and the patient recovered without complications. **Conclusion**: Anesthetic management in patients with suspected OI and mandibular neoplasms requires careful planning and execution. A multidisciplinary approach, including preoperative evaluation, airway management strategies, and close postoperative monitoring, is crucial for successful outcomes.

1. Introduction

Osteogenesis imperfecta (OI), often referred to as brittle bone disease, is a rare genetic disorder characterized by impaired collagen production, leading to fragile bones prone to fractures. The clinical presentation of OI varies widely, ranging from mild cases with few fractures to severe forms with numerous fractures, skeletal deformities, and significant disability. The estimated prevalence of OI is approximately 1 in 10,000 to 20,000 live births, highlighting its rarity and the unique challenges it presents in healthcare settings. The genetic mutations responsible for OI disrupt the synthesis or processing of type I collagen, the most abundant protein in the human body and a critical component of bone, skin, tendons, and other connective tissues. The resulting collagen deficiency compromises the structural integrity of these tissues, making them susceptible to fractures and deformities. OI is typically inherited in an autosomal dominant pattern, although autosomal recessive and sporadic cases have also been reported.[1,2]

The clinical manifestations of OI are diverse and can affect multiple organ systems. The hallmark of the disease is bone fragility, leading to frequent fractures, often occurring with minimal trauma or even spontaneously. These fractures can cause pain, deformity, and functional impairment. In addition to fractures, individuals with OI may experience other skeletal abnormalities, such as short stature,

scoliosis, bowing of long bones, and dental problems. Beyond the skeletal system, OI can also impact other tissues and organs. The sclerae, the white outer layer of the eyes, may appear blue or gray due to the underlying thinness and translucency of the collagen fibers. Hearing loss, often conductive in nature, can occur due to abnormalities in the middle ear bones. Dentinogenesis imperfecta, a condition affecting tooth development, may result in discolored, brittle teeth prone to wear and breakage. Respiratory complications, such as restrictive lung disease and recurrent pneumonia, can arise from chest wall deformities and muscle weakness. Cardiovascular manifestations, including aortic root dilatation and valvular insufficiency, have also been reported in some individuals with OI.[3,4]

The diagnosis of OI is typically based on a combination of clinical features, radiographic findings, and, in some cases, genetic testing. The Sillence classification system, which categorizes OI into four main types based on clinical severity and inheritance pattern, is commonly used to describe the phenotypic spectrum of the disease. Type I OI is the mildest form, characterized by blue sclerae, normal stature, and few fractures. Type II OI is the most severe form, often lethal in the perinatal period due to multiple fractures and respiratory insufficiency. Types III and IV OI represent intermediate forms with varying degrees of bone fragility and skeletal deformities. The management of OI is complex and requires a multidisciplinary approach involving various healthcare professionals, including orthopedic surgeons, geneticists, physical therapists, and anesthesiologists. The primary goals of treatment are to prevent fractures, minimize deformities, and optimize function. This may involve a combination of non-surgical and surgical interventions.[5,6]

Non-surgical management focuses on promoting bone health and preventing fractures through lifestyle modifications, physical therapy, and medications. Adequate calcium and vitamin D intake, weight-bearing exercises, and fall prevention strategies are crucial for maintaining bone strength and reducing fracture risk. Bisphosphonates, medications that inhibit bone resorption, have been shown to increase bone mineral density and reduce fracture rates in some individuals with OI. Surgical interventions may be necessary to correct deformities, stabilize fractures, or improve function. Intramedullary rodding, a procedure involving the insertion of metal rods into the medullary cavity of long bones, can provide stability and prevent fractures. Osteotomies, surgical cuts in the bone, may be performed to correct angular deformities or limb length discrepancies. Spinal fusion surgery may be indicated for severe scoliosis. Anesthetic management of patients with OI presents unique challenges due to the inherent fragility of their bones and the potential for other comorbidities. Careful handling and positioning are essential to avoid fractures during transport and surgery. Airway management can be difficult due to potential cervical spine instability, facial deformities, and limited mouth opening. Intubation may require specialized techniques, such as fiberoptic bronchoscopy or awake intubation. The choice of anesthetic agents and techniques should be tailored to the individual patient, considering their specific needs and comorbidities.[7,8]

In addition to the challenges associated with OI, the presence of a mandibular neoplasm further complicates anesthetic management. Mandibular neoplasms can cause significant distortion of the anatomy, making airway access and surgical resection more challenging. The tumor may also compromise the patient's ability to open their mouth, further limiting airway options. Moreover, depending on the type and extent of the tumor, there may be concerns about bleeding, airway obstruction, or nerve injury during surgery.[9,10] The case presented in this report highlights the successful anesthetic management of a patient with a mandibular neoplasm and suspected OI. The patient's clinical presentation, including short stature, bone deformities, and abnormal tooth growth, raised suspicion for OI, although genetic confirmation was not available. The large mandibular tumor caused significant airway obstruction and necessitated a

tracheostomy for safe airway management. The anesthetic team carefully planned and executed the procedure, considering the patient's unique needs and potential complications.

2. Case Presentation

A 33-year-old male patient presented to the hospital with a chief complaint of a progressively enlarging mass in his oral cavity. The mass, initially the size of a marble, had been present for one year but had rapidly grown over the past four months, leading to complete obstruction of his mouth. This obstruction severely impacted his quality of life, causing difficulty breathing, particularly when lying supine, and necessitating reliance on a nasogastric tube for feeding. The patient's medical history was significant for multiple fractures and skeletal deformities since childhood, raising suspicion for an underlying diagnosis of osteogenesis imperfecta (OI). He had no prior surgeries or significant medical conditions apart from his current presentation.

On physical examination, the patient exhibited characteristic features suggestive of OI. His height was markedly short at 90 cm, and his weight was 35 kg, indicating growth retardation. Notably, all of his extremities displayed deformities, further supporting the suspected diagnosis. Examination of the head and neck region revealed a short and rigid neck with a limited range of motion. The oral cavity was entirely occupied by a large tumor mass, precluding visualization of the oropharynx and making traditional airway assessment impossible. The patient's sclerae were normal in color, and he reported no hearing impairments. However, he did note a history of abnormal tooth growth prior to the development of the tumor. A comprehensive physical examination of the cardiovascular and respiratory systems was unremarkable, suggesting no significant comorbidities in these areas.

Preoperative laboratory investigations were conducted to assess the patient's overall health status and identify any potential concerns that might impact anesthetic management. The results of these investigations were within normal limits, including a hemoglobin level of 12.2 mg/dL, indicating no significant anemia. Electrocardiography (ECG) also showed no abnormalities, suggesting normal cardiac function. Imaging studies played a crucial role in characterizing the extent of the patient's condition. A bone survey revealed generalized osteopenia, a decrease in bone density, and evidence of previous fractures, further supporting the suspected diagnosis of OI. A computed tomography (CT) scan of the neck provided detailed visualization of the tumor, revealing a large, heterogeneous mass in the right mandible with areas of necrosis. These findings were highly suspicious of malignancy. The CT scan also demonstrated significant destruction of the mandibular bone and the presence of bilateral cervical lymphadenopathy, indicating the potential spread of the tumor to regional lymph nodes.

The combination of the patient's clinical presentation, physical examination findings, and imaging results led to a working diagnosis of a mandibular neoplasm in the setting of suspected OI. The patient's history of multiple fractures, skeletal deformities, and abnormal tooth growth, along with the radiographic evidence of osteopenia and previous fractures, strongly suggested OI. While genetic testing was not available to confirm the diagnosis definitively, the clinical picture was highly suggestive. The large, heterogeneous mandibular mass with areas of necrosis seen on the CT scan raised significant concern for malignancy. The extensive destruction of the mandible and the presence of bilateral cervical lymphadenopathy further underscored the aggressive nature of the tumor.

The primary treatment goal was surgical resection of the mandibular neoplasm. However, the presence of suspected OI and the challenging anatomical location of the tumor necessitated careful planning and a multidisciplinary approach. The anticipated difficult airway posed a significant challenge for anesthetic management. The tumor's complete obstruction of the oral cavity, combined with the patient's short neck and limited neck mobility, made traditional intubation

techniques impractical and potentially dangerous. Therefore, the decision was made to secure the airway through an ultrasound-guided tracheostomy under local anesthesia before proceeding with general anesthesia and surgical resection. The surgical plan involved complete resection of the tumor with adequate margins to ensure complete removal of malignant tissue. Reconstruction of the mandible was also planned to restore function and aesthetics.

The patient's prognosis was guarded due to the suspected malignancy of the tumor and the potential complications associated with OI. However, with successful surgical resection and appropriate postoperative care, the outlook for long-term survival and functional recovery was favorable. Close follow-up was planned to monitor for recurrence of the tumor and manage any complications related to OI or the surgical procedure. Regular imaging studies and clinical examinations would be essential for ongoing surveillance. This case presentation highlights the complex interplay between a rare genetic disorder and a challenging surgical condition. The successful management of this patient underscores the importance of a multidisciplinary approach, careful planning, and individualized anesthetic care in achieving optimal outcomes for patients with unique and complex medical needs.

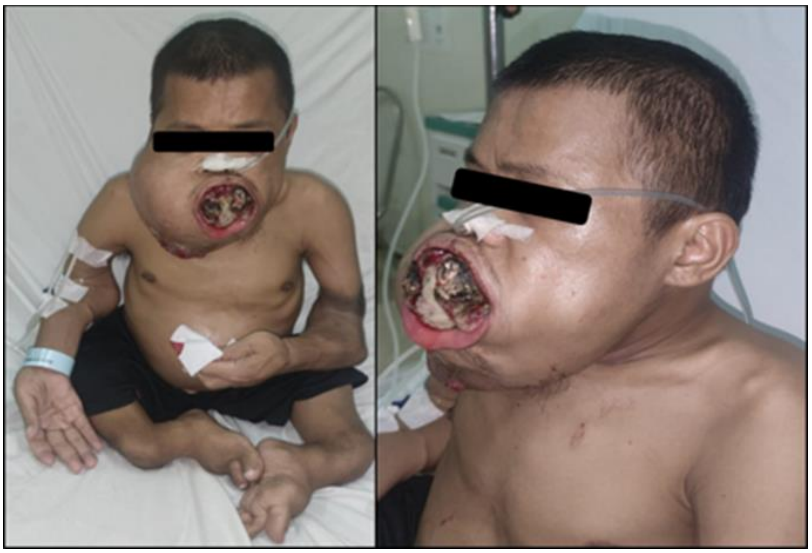


Figure 1. Clinical photo of the patient.

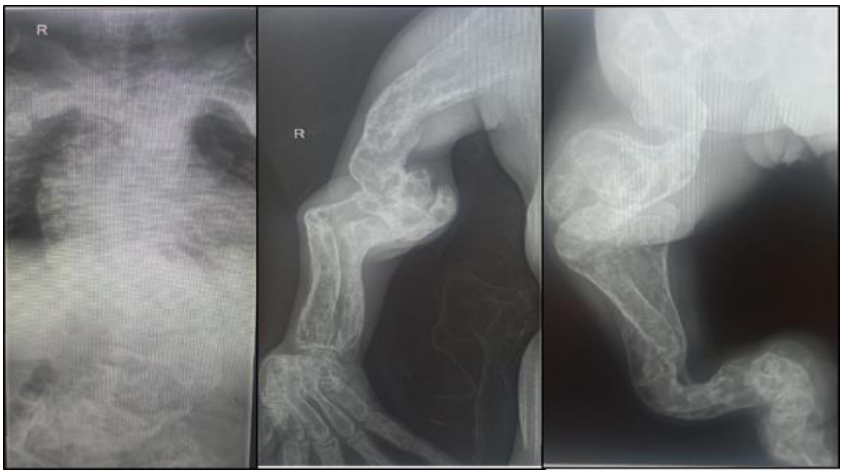


Figure 2. Rontgen bone survey: osteogenesis imperfecta.

3. Discussion

Osteogenesis imperfecta (OI), also known as brittle bone disease, is a complex and multifaceted genetic disorder primarily characterized by bone fragility and a predisposition to fractures. The condition arises from defects in the production or processing of type I collagen, a vital structural protein that provides strength and resilience to various connective tissues throughout the body, including bone, skin, tendons, and ligaments. The clinical manifestations of OI are remarkably diverse, spanning a spectrum from mild cases with relatively few fractures to severe forms associated with numerous fractures, significant skeletal deformities, and substantial functional impairment. At its core, OI is a disorder of collagen, the most abundant protein in the human body. Collagen forms the structural framework of many tissues, providing them with tensile strength and elasticity. Type I collagen, the specific type affected in OI, is a major component of bone, skin, tendons, ligaments, and other connective tissues. The genetic mutations responsible for OI disrupt the normal synthesis or processing of type I collagen. These mutations can occur in various genes involved in collagen production, including COL1A1 and COL1A2, which encode the two chains of the type I collagen molecule. The resulting collagen deficiency or structural abnormalities compromise the integrity of connective tissues, making them susceptible to fractures and deformities. OI is typically inherited in an autosomal dominant pattern, meaning that a single copy of the mutated gene is sufficient to cause the disorder. However, autosomal recessive and sporadic cases have also been reported, adding to the genetic complexity of OI. The clinical presentation of OI is remarkably heterogeneous, reflecting the wide range of genetic mutations and their impact on collagen production. The hallmark of the disease is bone fragility, leading to frequent fractures, often occurring with minimal trauma or even spontaneously. These fractures can cause pain, deformity, and functional impairment, significantly impacting the quality of life of affected individuals. In addition to fractures, individuals with OI may experience a variety of other skeletal abnormalities. Short stature is a common feature, particularly in more severe forms of the disease. Scoliosis, an abnormal curvature of the spine, can also occur, potentially leading to respiratory compromise and other complications. Bowing of long bones, such as the legs, is another characteristic finding, often contributing to gait abnormalities and functional limitations. Dental problems, including dentinogenesis imperfecta, a condition affecting tooth development, may result in discolored, brittle teeth prone to wear and breakage. Beyond the skeletal system, OI can also impact other tissues and organs. The sclerae, the white outer layer of the eyes, may appear blue or gray due to the underlying thinness and translucency of the collagen fibers. This is a classic sign of OI, although it is not present in all cases. Hearing loss, often conductive in nature, can occur due to abnormalities in the middle ear bones, specifically the ossicles, which are responsible for transmitting sound vibrations. Respiratory complications can arise from chest wall deformities and muscle weakness associated with OI. Restrictive lung disease, a condition characterized by reduced lung volume and decreased lung compliance, can impair respiratory function and lead to exercise intolerance and shortness of breath. Recurrent pneumonia may also occur due to weakened respiratory muscles and impaired clearance of secretions. Cardiovascular manifestations, although less common, have also been reported in some individuals with OI. Aortic root dilatation, an enlargement of the first part of the aorta, can increase the risk of aortic dissection or rupture, a life-threatening condition. Valvular insufficiency, particularly affecting the mitral and aortic valves, can lead to heart failure and other complications. The diagnosis of OI is typically based on a combination of clinical features, radiographic findings, and, in some cases, genetic testing. A detailed medical history and physical examination are essential for identifying characteristic features of OI, such as a history of multiple fractures, skeletal deformities, blue sclerae,

and dental abnormalities. Radiographic imaging, including X-rays and bone scans, can reveal evidence of fractures, osteopenia (decreased bone density), and other skeletal abnormalities associated with OI. These findings can help confirm the diagnosis and assess the severity of the disease. Genetic testing, although not always necessary for diagnosis, can be helpful in confirming the specific genetic mutation responsible for OI. This information can be valuable for family planning and predicting the potential severity of the disease in future generations. The Sillence classification system, developed in the 1970s, is widely used to categorize OI into four main types based on clinical severity and inheritance pattern. Type I OI is the mildest form, characterized by blue sclerae, normal stature, and relatively few fractures. Individuals with type I OI typically have a normal lifespan and experience minimal functional impairment. Type II OI is the most severe form, often lethal in the perinatal period or early infancy due to multiple fractures, respiratory insufficiency, and other complications. Infants with type II OI often have a characteristic appearance with short limbs, a soft skull, and multiple fractures visible on prenatal ultrasound. Type III OI is an intermediate form characterized by progressive skeletal deformities, short stature, and multiple fractures. Individuals with type III OI often require assistive devices for mobility and may experience significant functional limitations. Type IV OI is another intermediate form with varying degrees of bone fragility and skeletal deformities. Individuals with type IV OI may have normal or gray sclerae and typically experience fewer fractures than those with type III OI. The management of OI is complex and requires a multidisciplinary approach involving various healthcare professionals, including orthopedic surgeons, geneticists, physical therapists, occupational therapists, dentists, audiologists, and anesthesiologists. The primary goals of treatment are to prevent fractures, minimize deformities, and optimize function, thereby enhancing the quality of life for individuals with OI. Non-surgical management focuses on promoting bone health and preventing fractures through lifestyle modifications, physical therapy, and medications. Adequate calcium and vitamin D intake is crucial for maintaining bone strength and reducing fracture risk. Weight-bearing exercises, such as walking and swimming, can help improve bone density and muscle strength. Fall prevention strategies, including home modifications and assistive devices, are important for minimizing the risk of fractures. Bisphosphonates, medications that inhibit bone resorption, have been shown to increase bone mineral density and reduce fracture rates in some individuals with OI, particularly children and adolescents. However, the long-term benefits and potential side effects of bisphosphonate therapy remain areas of active research. Surgical interventions may be necessary to correct deformities, stabilize fractures, or improve function. Intramedullary rodding, a procedure involving the insertion of metal rods into the medullary cavity of long bones, can provide stability and prevent fractures, particularly in the lower extremities. Osteotomies, surgical cuts in the bone, may be performed to correct angular deformities or limb length discrepancies. Spinal fusion surgery may be indicated for severe scoliosis to prevent further progression and improve spinal stability. Dental care is an important aspect of OI management, as individuals with dentinogenesis imperfecta may require specialized dental treatments to address tooth fragility and wear. Regular dental checkups, preventive care, and restorative procedures can help maintain oral health and function. Hearing aids or other assistive listening devices may be beneficial for individuals with OI who experience hearing loss. Audiological evaluations should be performed regularly to monitor hearing function and identify any changes that may require intervention. Anesthetic management of patients with OI presents unique challenges due to the inherent fragility of their bones and potential for other comorbidities. Careful handling and positioning are essential to avoid fractures during transport and surgery. Airway management can be difficult due to potential cervical spine instability, facial deformities, and limited mouth

opening. Intubation may require specialized techniques, such as fiberoptic bronchoscopy or awake intubation, to minimize the risk of trauma and complications. The choice of anesthetic agents and techniques should be tailored to the individual patient, considering their specific needs and comorbidities. Inhalational anesthetics, such as sevoflurane and desflurane, are generally considered safe for patients with OI, although careful titration and monitoring are necessary to avoid hypotension and respiratory depression. Intravenous anesthetics, such as propofol and remifentanil, can also be used, but caution should be exercised in patients with cardiovascular or respiratory compromise. Muscle relaxants should be used judiciously in patients with OI, as they can increase the risk of fractures and respiratory complications. Careful monitoring of neuromuscular function and the use of reversal agents when necessary are important for ensuring adequate respiratory function and minimizing the risk of postoperative complications. Pain management is a critical aspect of postoperative care for patients with OI. Opioids, nonsteroidal anti-inflammatory drugs (NSAIDs), and other analgesics can be used to control pain, but careful titration and monitoring are necessary to avoid respiratory depression and other side effects. Regional anesthesia techniques, such as nerve blocks, may also be beneficial for providing effective pain relief while minimizing the need for systemic opioids.[11-13]

Anesthetic management of patients with osteogenesis imperfecta (OI) necessitates a meticulous and well-informed approach due to the inherent fragility of their bones and the potential presence of various comorbidities. The primary concerns in providing anesthesia to these patients revolve around airway management, fracture risk, bleeding tendencies, hyperthermia, and cardiac abnormalities. Each of these aspects requires careful consideration and proactive management to ensure patient safety and optimal outcomes. Securing and maintaining a patent airway in patients with OI can be particularly challenging due to a confluence of factors. One of the primary concerns is the potential for cervical spine instability, a complication that can arise from the underlying collagen defect in OI. This instability can render intubation and positioning maneuvers more difficult and increase the risk of iatrogenic spinal cord injury. Consequently, anesthesiologists must exercise extreme caution and employ techniques that minimize neck movement and manipulation. Facial deformities, such as midface hypoplasia (underdevelopment of the midface) and micrognathia (small jaw), can further complicate airway access in patients with OI. These deformities may alter the normal anatomical landmarks and relationships, making it challenging to visualize the glottis and insert an endotracheal tube. In some cases, the presence of these deformities may necessitate the use of alternative airway management techniques, such as fiberoptic intubation or laryngeal mask airway. Limited mouth opening, another potential challenge in OI, can arise from temporomandibular joint involvement or muscle contractures. This limitation can significantly impede access to the airway and make conventional laryngoscopy difficult. Anesthesiologists may need to employ specialized techniques, such as awake fiberoptic intubation or the use of a flexible laryngoscope, to overcome this obstacle. The hallmark of OI is bone fragility, rendering patients highly susceptible to fractures even with minimal manipulation or stress. This vulnerability necessitates meticulous care and attention to detail throughout the perioperative period. Careful handling and positioning are paramount to avoid iatrogenic fractures. Gentle padding and support should be used to protect bony prominences, and movements should be minimized to avoid unnecessary stress on the skeletal system. During induction of anesthesia and transfer to the operating table, particular care must be taken to avoid sudden or forceful movements. The use of assistive devices, such as slide boards or transfer sheets, can help facilitate safe and gentle patient movement. Intraoperatively, positioning should be optimized to ensure patient comfort and stability while minimizing pressure on vulnerable areas. The use of specialized

positioning aids, such as gel pads and bean bags, can help distribute pressure evenly and reduce the risk of fractures. Although not observed in the specific case presented in this report, patients with OI may exhibit a bleeding tendency due to platelet dysfunction or abnormalities in coagulation factors. This predisposition to bleeding can complicate surgical procedures and increase the risk of perioperative hemorrhage. Preoperative assessment of coagulation status, including a thorough review of the patient's medical history and laboratory investigations such as platelet count, prothrombin time, and activated partial thromboplastin time, is crucial for identifying any potential bleeding risks. If abnormalities are detected, appropriate measures, such as platelet transfusion or administration of coagulation factors, may be necessary to optimize hemostasis. During surgery, meticulous surgical technique and the use of hemostatic agents can help minimize blood loss. Anesthesiologists should be vigilant in monitoring blood loss and be prepared to administer blood products or other interventions if necessary to maintain adequate intravascular volume and oxygen-carrying capacity. Individuals with OI may be prone to hyperthermia, an elevation of core body temperature above the normal range. This predisposition can be attributed to an increased metabolic rate or impaired thermoregulation associated with the underlying collagen defect. Hyperthermia can have detrimental effects on various organ systems and increase the risk of complications during and after surgery. Careful monitoring of core body temperature is essential throughout the perioperative period. Measures to prevent hyperthermia, such as avoiding excessive warming and using cool fluids for intravenous administration and irrigation, should be implemented. If hyperthermia occurs, prompt intervention with cooling measures, such as ice packs, cooling blankets, and chilled intravenous fluids, is necessary to restore normothermia and prevent complications. Some patients with OI may have cardiac abnormalities, such as aortic root dilatation or valvular insufficiency, which can impact anesthetic management. Aortic root dilatation, an enlargement of the first part of the aorta, can increase the risk of aortic dissection or rupture, a life-threatening complication. Valvular insufficiency, particularly affecting the mitral and aortic valves, can lead to heart failure and other cardiovascular complications. Preoperative echocardiography may be indicated in selected cases to assess cardiac function and identify any potential concerns that may influence anesthetic management. If significant cardiac abnormalities are detected, consultation with a cardiologist may be warranted to optimize medical management and minimize perioperative risks.[14-16]

The presence of a mandibular neoplasm in a patient with osteogenesis imperfecta (OI) introduces a significant layer of complexity to anesthetic management, demanding heightened vigilance and adaptability from the healthcare team. Mandibular tumors, whether benign or malignant, can exert a profound impact on the anatomical landscape of the oral cavity and surrounding structures, creating formidable challenges for airway access, surgical resection, and overall anesthetic care. Mandibular neoplasms, by their very nature, can cause substantial distortion of the normal anatomy of the jaw, oral cavity, and adjacent tissues. This distortion can manifest in various ways, depending on the size, location, and growth pattern of the tumor. Large tumors may displace or compress vital structures, such as the tongue, pharynx, and larynx, potentially compromising the airway and making intubation challenging. Infiltrative tumors may invade surrounding tissues, including muscles and nerves, further complicating surgical access and increasing the risk of complications. The presence of a mandibular tumor can also significantly limit the patient's ability to open their mouth, a phenomenon known as trismus. Trismus can arise from several factors, including tumor infiltration of the muscles of mastication, inflammation, or pain. This limitation in mouth opening can severely restrict access to the airway, making conventional laryngoscopy and intubation difficult or even impossible. In the context of OI, where airway management is already

challenging due to potential cervical spine instability and facial deformities, the presence of a mandibular neoplasm amplifies these difficulties. Anesthesiologists must carefully assess the extent of anatomical distortion and anticipate potential airway challenges to develop a safe and effective airway management plan. The surgical resection of a mandibular neoplasm is a complex procedure that requires meticulous planning and execution. The presence of OI adds another layer of complexity, as the surgeon must navigate the fragile bone and surrounding tissues with utmost care to avoid fractures and other complications. The extent of the tumor and its involvement of critical structures, such as nerves and blood vessels, will dictate the surgical approach and the potential for complications. Large tumors may necessitate extensive resection of the mandible, potentially leading to significant functional and aesthetic deficits. Reconstruction of the mandible may be required to restore form and function, further adding to the complexity of the procedure. In patients with OI, the risk of intraoperative fractures is heightened due to the inherent fragility of their bones. The surgeon must exercise extreme caution during tumor resection and reconstruction to avoid excessive force or manipulation that could lead to fractures. The use of specialized instruments and techniques may be necessary to minimize the risk of iatrogenic injury. Mandibular neoplasms, particularly those that are large or invasive, carry a risk of various complications that can impact anesthetic management. Bleeding is a major concern, especially in the context of OI, where patients may have a predisposition to bleeding due to platelet dysfunction or coagulation abnormalities. The surgical team must be prepared to manage significant blood loss and have blood products readily available for transfusion if necessary. Airway obstruction is another potential complication, particularly during tumor resection or manipulation of the airway. The anesthesiologist must be vigilant in monitoring the patient's airway and be prepared to intervene promptly if obstruction occurs. This may involve repositioning the patient, suctioning the airway, or, in severe cases, re-establishing the airway through emergency tracheostomy or cricothyrotomy. Nerve injury is a risk associated with any surgical procedure in the head and neck region. In the context of mandibular neoplasms, the inferior alveolar nerve, which provides sensation to the lower lip and chin, is particularly vulnerable to injury. The surgeon must take great care to identify and preserve this nerve during tumor resection. However, if nerve injury does occur, it can lead to numbness or paresthesia in the affected area. The anesthetic management of patients with OI and mandibular neoplasms requires a multidisciplinary approach and careful planning. The anesthesiologist, surgeon, and other members of the healthcare team must collaborate closely to develop a comprehensive plan that addresses the unique needs and challenges of each patient. Preoperative evaluation is crucial for identifying potential risks and developing an appropriate anesthetic plan. This evaluation should include a thorough assessment of the airway, cervical spine, and other comorbidities. Imaging studies, such as CT scans and MRIs, can provide valuable information about the extent of the tumor and its impact on surrounding structures. Airway management strategies should be tailored to the individual patient, taking into account the size and location of the tumor, the degree of trismus, and the potential for cervical spine instability. In cases where conventional intubation is not feasible, alternative techniques, such as fiberoptic intubation, laryngeal mask airway, or tracheostomy, may be necessary. Intraoperative monitoring and management should focus on maintaining hemodynamic stability, ensuring adequate oxygenation and ventilation, and preventing complications such as fractures, bleeding, and nerve injury. Careful positioning, gentle handling, and meticulous surgical technique are essential for minimizing the risk of iatrogenic complications. Postoperative care should include close monitoring for complications, such as bleeding, airway obstruction, and infection. Pain management is also crucial, as patients with OI may experience significant pain due to fractures or surgical trauma. A multimodal

approach to pain management, including opioids, NSAIDs, and regional anesthesia techniques, may be necessary to provide adequate analgesia while minimizing side effects.[17,18]

The case presented in this report exemplifies the intricate challenges encountered when managing a patient with a mandibular neoplasm in the context of suspected osteogenesis imperfecta (OI). The confluence of these two conditions creates a unique clinical scenario that demands meticulous planning, adaptability, and a multidisciplinary approach to ensure patient safety and successful surgical and anesthetic outcomes. The patient's large mandibular tumor presented a formidable obstacle to airway management. The tumor's complete obstruction of the oral cavity, coupled with the patient's short neck and limited neck mobility due to suspected OI, rendered traditional intubation techniques not only impractical but also potentially hazardous. Attempting laryngoscopy and intubation in such a scenario could lead to significant trauma, fractures, and even airway compromise. Recognizing the anticipated difficult airway, the anesthetic team opted for a proactive and innovative approach: an ultrasound-guided tracheostomy performed under local anesthesia. This decision was pivotal in ensuring secure airway control and ventilation throughout the surgical procedure. Ultrasound guidance facilitated precise needle placement and incision, minimizing the risk of complications and optimizing the success of the tracheostomy. Performing the tracheostomy under local anesthesia allowed for airway control without the need for general anesthesia and its associated risks in this high-risk patient. The inherent bone fragility associated with OI necessitates meticulous care and attention to detail throughout the perioperative period. Even minor manipulations or movements can precipitate fractures in these patients, underscoring the importance of gentle handling and strategic positioning. In this case, the anesthetic team implemented several measures to mitigate fracture risk. The patient was carefully transported to the operating room, ensuring minimal movement and avoiding any sudden or jarring motions. During positioning on the operating table, gentle padding and support were used to protect bony prominences, and the patient's limbs were carefully aligned to avoid undue stress on the joints. Throughout the surgical procedure, movements were kept to a minimum, and the surgical team exercised utmost caution during the manipulation of the patient's head and neck. While the patient in this case did not exhibit any overt bleeding tendencies during surgery, the potential for hemorrhage remained a significant concern. OI can be associated with platelet dysfunction or abnormalities in coagulation factors, increasing the risk of perioperative bleeding. The anesthetic team took proactive measures to address this potential complication. Preoperative coagulation studies were performed to assess the patient's bleeding risk, and blood products were readily available for transfusion if necessary. During surgery, meticulous surgical technique and the use of hemostatic agents helped minimize blood loss. The anesthesiologist remained vigilant in monitoring blood loss and was prepared to administer blood products or other interventions if needed to maintain hemodynamic stability. Patients with OI may be prone to hyperthermia due to an increased metabolic rate or impaired thermoregulation. Hyperthermia can have detrimental effects on various organ systems and increase the risk of complications during and after surgery. In this case, the anesthetic team closely monitored the patient's core body temperature throughout the procedure. Measures to prevent hyperthermia, such as avoiding excessive warming and using cool fluids for intravenous administration and irrigation, were implemented. The operating room temperature was also adjusted to maintain a comfortable environment for the patient. These proactive measures successfully prevented any significant temperature elevations, ensuring normothermia throughout the surgical procedure. This case report serves as a powerful reminder of the multifaceted challenges involved in managing patients with OI undergoing surgical procedures, particularly in the context of complex

conditions such as mandibular neoplasms. The successful outcome in this case underscores the importance of a multidisciplinary approach, meticulous planning, and individualized anesthetic care. The decision to perform an ultrasound-guided tracheostomy under local anesthesia proved to be a critical turning point in this case, allowing for safe and effective airway management in the face of a seemingly insurmountable obstacle. This innovative approach highlights the importance of adaptability and the willingness to explore alternative techniques when traditional methods are not feasible or safe. The careful attention to fracture risk mitigation, hemorrhage preparedness, and temperature regulation further demonstrates the comprehensive and proactive approach required in the anesthetic management of patients with OI. By anticipating potential complications and implementing appropriate preventive measures, the anesthetic team was able to minimize risks and optimize patient outcomes. This case also emphasizes the importance of collaboration and communication among the various healthcare professionals involved in the care of patients with OI. The anesthesiologist, surgeon, and other members of the team must work together seamlessly to develop and execute a comprehensive plan that addresses the unique needs and challenges of each patient. [19,20]

4. Conclusion

The successful anesthetic management of a patient with a mandibular neoplasm and suspected osteogenesis imperfecta (OI) underscores the importance of a thorough preoperative assessment, careful planning, and a multidisciplinary approach. The presence of OI necessitates meticulous attention to airway management, fracture risk mitigation, and potential bleeding complications. The use of ultrasound-guided tracheostomy under local anesthesia proved to be a safe and effective strategy for securing the airway in this challenging case. This report emphasizes the need for individualized anesthetic care and highlights the potential for successful outcomes even in complex clinical scenarios.

5. References

1. Chang CP, Abdelwahab M, Liu SY. Maxillomandibular advancement followed by upper airway stimulation as long-term management of obstructive sleep apnea. J Oral Maxillofac Surg. 2022; 80(9): S29–30.
2. Zhou Y, Wang X, Li Z, Yang X. Airway management under general anesthesia for infants and young children with the first and second branchial syndrome featuring unilateral mandibular dysplasia: a case series of 8 children. J Stomatol Oral Maxillofac Surg. 2023; 124(6S2): 101558.
3. Shah SV, Chaggar RS. Advanced airway management techniques in anaesthesia for oral cancer surgery: a review. J Oral Maxillofac Anesth. 2023; 2: 8–8.
4. Dahiya A, Singh V, Bhagol A, Britto A, Chhikara D. Lunch box penetrating injury in the craniofacial region impedes the primary airway management and surgical intervention. J Maxillofac Oral Surg. 2023; 22(2): 460–3.
5. Vyas SR, Jain S, Mogra G, Kela G. Modified endotracheal tube for airway management in paediatric patients with Pierre Robin Sequence. Br J Oral Maxillofac Surg. 2024; 62(2): 213–4.
6. Malladi UK. Airway management in Ludwig's angina: a narrative review. Oral Oncology Reports. 2024; 10(100312): 100312.
7. Mukherjee S, Jain A, S S, Waindeskar V. Clinical outcome of perioperative airway and ventilatory management in patients undergoing surgery for oral cavity cancer: a prospective observational study. J Korean Assoc Oral Maxillofac Surg. 2024; 50(3): 146–52.
8. Gupta A, Poorna TA, Khismatrao VN. Can prophylactic tracheostomy be avoided in advanced oral cancer surgery for airway

management? An experience and guidelines from a tertiary care facility. J Maxillofac Oral Surg. 2024; 23(4): 918–22.
9. Merola R, Troise S, Palumbo D, D'Auria D, Dell'Aversana Orabona G, Vargas M. Airway management in patients undergoing maxillofacial surgery: State of art review. J Stomatol Oral Maxillofac Surg. 2024; (102044): 102044.
10. Rodrigues MM. Correlation between the Friedman staging system and the upper airway volume in patients with obstructive sleep apnea. Oral Health Dent Manag. 2016; 15(07).
11. Zotarelli Filho IJ, Kassis EN, Marques DC, Tosto Dias LF, Albuquerque Barros A, Luiz de AraAAjo Vian R, et al. Radiographic analysis of posterior airway variation, gonial angle, mandibular length and facial height in patients submitted to vertical mandible osteotomy. OHDM. 2019; 18(1).
12. Liu WJ, Gong LG, Tan CM, Li QY, Ma YZ. McCune-Albright syndrome with initial symptom of mandibular neoplasms: a case report. Zhonghua Er Bi Yan Hou Tou Jing Wai Ke Za Zhi. 2017; 52(2): 122–4.
13. Peacock ZS, Ji YD. Cost and lack of insurance coverage are prohibitive to having dental implants after resections for benign mandibular neoplasms. Oral Surg Oral Med Oral Pathol Oral Radiol. 2017; 123(6): 645–50.
14. Radkevich AA, Gantimurov AA, Gunther VE. Elimination of mandibular defects in patients with malignant neoplasms using the poprus TiNi-alloys. KnE Mater Sci. 2017; 2(1): 286.
15. Friedrich RE, Chin S-J, Luebke AM, Speth US, Assaf AT, Gosau M. A connective tissue neoplasm of the mandibular angle mimicking Stafne's bone cavity on panoramic view. Anticancer Res. 2021; 41(8): 3983–8.
16. Gabriele G, Funaioli F, Cascino F, Grandini S, Fantozzi V, Gennaro P. A rare mandibular neoplasm: case report of a Central Giant Cell Granuloma. Ann Ital Chir. 2021; 92.
17. Vishal, Rohit, Prajapati VK, Shahi AK, Prakash O. Evaluation of functional and esthetic outcome of patients after reconstruction with mandibular reconstruction plates preceded by resection of benign odontogenic neoplasms of mandible: a cohort study. Indian J Otolaryngol Head Neck Surg. 2022; 74(Suppl 2): 2350–5.
18. Nieborak R, Rolski D. Implantoprosthetic rehabilitation of a patient after mandibular resection due to neoplasm and reconstruction with microvascular free fibular flap – a case report. Protet Stomatol. 2022; 72(4): 369–74.
19. Ozel MA, Ogul H. Osteoid osteoma: an unusual neoplasm of the mandibular condyle. Br J Hosp Med (Lond). 2022; 83(5): 1.
20. Isaias PHC, Amorim PHGH, Costa FWG, Pereira KMA, Sousa FB, Alves APNN, et al. Extensive mandibular lesion as a primary sign of neoplasm of plasma cell origin: a case report. Oral Surg Oral Med Oral Pathol Oral Radiol. 2023; 136(1): e27.