

Case Report of a Difficult Airway due to Calcinosis Cutis Universalis: An Anaesthesiologist Perspective

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ABSTRACT

Difficult airway is defined as a situation where a trained anaesthesiologist encounters challenges in either face mask ventilation, tracheal intubation, or both, as stated by the American Society of Anaesthesiologists. A case of a difficult airway due to Calcinosis Universalis was encountered, which is a rare subtype of connective tissue disorder. In Calcinosis Cutis, calcium is diffusely deposited in the skin, subcutaneous tissue, muscles, and tendons. Calcinosis Universalis typically appears secondary to a connective tissue disorder such as juvenile dermatomyositis, systemic lupus erythematosus, Sjögren's syndrome, and Calcinosis, Raynaud's phenomenon, Esophageal dysmotility, Sclerodactyly, and Telangiectasia (CREST) syndrome, predominantly affecting females. There is no specific treatment available for this condition. Medically, it can be managed with calcium channel blockers like diltiazem, colchicine, immunosuppressants, and painkillers to alleviate discomfort. Antibiotics may be required for infected wounds. Surgically, lesions can be removed; however, there is a higher likelihood of recurrence. Patients with Calcinosis Universalis present a challenge for anaesthesiologists due to the involvement of joints and tendons. The present case report involves a 22-year-old female with a history of juvenile dermatomyositis for the past 15 years and Calcinosis Cutis universalis for the past 11 years presented to the Anaesthesiology department for nasal bone reduction. Upon examination, it was determined that she had a difficult airway, and awake fiberoptic intubation was performed. The present case report aims to provide unique insights into a rare case, highlighting the unusual presentation of a difficult airway and the associated perioperative challenges from an anaesthesiologist's perspective.

Keywords: Awake intubation, Autoimmune connective tissue disorder, CREST syndrome, Difficult airway, Diltiazem, Juvenile dermatomyositis

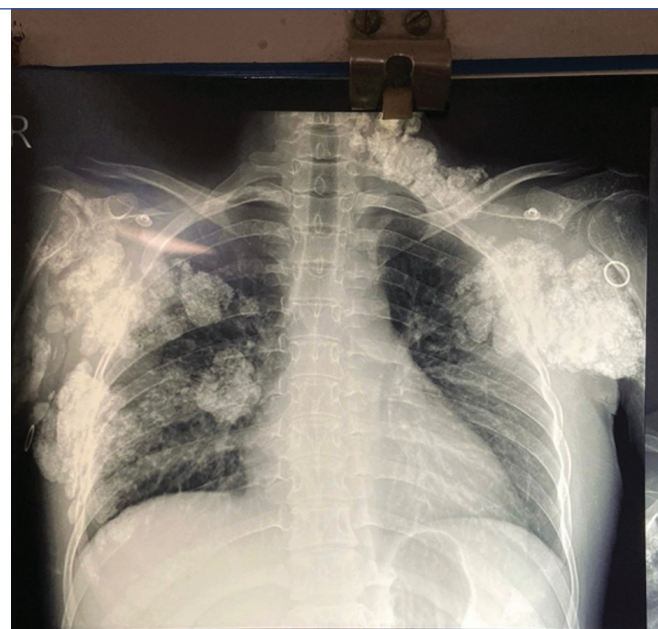
CASE REPORT

A 22-year-old female presented to the Oral and Maxillofacial Surgery Department with an alleged history of a slip and fall a week ago, resulting in an injury to her nose. A history of nosebleed was noted acutely after the fall, but it resolved subsequently. She has been a known case of juvenile dermatomyositis since the age of seven and was diagnosed with Calcinosis Cutis universalis at the age of 11. For this condition, she was treated with Tab. Diltiazem 60 mg BD and Tab. Colchicine 0.5 mg BD.

On general examination, multiple lesions were observed over the temporomandibular joints, the anterior aspect of the neck on the left side, as well as the elbows and wrists. The largest lesion measured approximately 4×3 cm, while the smallest measured about 1×1 cm and appeared hard in consistency. There was no pallor, icterus, clubbing, cyanosis, or pedal edema. Her blood pressure was 120/90 mmHg, and her pulse rate was 86 beats per minute.

Upon examining her neck, it was found to have restricted extension, with mouth opening limited to only one and a half fingers vertically. Her Mallampati score could not be assessed due to restricted mouth opening, but the thyromental distance was normal. Further evaluation included a complete blood count, renal function tests, liver function tests, serum electrolytes, coagulation profile, chest X-ray, and electrocardiogram. All lab investigations were normal, and the chest X-ray revealed multiple calcific sites on the left side of the neck, both axillae, and the chest region [Table/Fig-1].

Nasal bone reduction surgery was planned, for which the patient came to the Anaesthesiology department. The mode of anaesthesia and the associated risks were explained to her. Informed and written consent was obtained, and premedications consisting of Tab. Alprazolam 0.25 mg, Tab. Metoclopramide 10 mg, and Tab. Ranitidine 150 mg were prescribed before bedtime and on the



[Table/Fig-1]: Chest X-ray posteroanterior view of the patient showing multiple lesions of calcinosis.

morning of the surgery. Given the difficulty of the airway, we planned for awake fiberoptic intubation. The patient was nebulised with 4% lignocaine (1 mL), and nasal packing was performed with 2 mL of 4% lignocaine in each nostril for 10 minutes in the premedication room.

The patient was then shifted to the operating theater, where monitors were attached. An oropharyngeal spray with 1% lignocaine was administered. An airway block was given, consisting of 2 mL of 1% lignocaine for a bilateral superior laryngeal nerve block using

an external approach by piercing the thyrohyoid membrane near the greater cornu of the hyoid, and 2 mL of 4% lignocaine for a recurrent laryngeal nerve block using a translaryngeal approach by piercing the cricothyroid membrane, all under aseptic precautions. The patient was intubated with a 6.5 mm cuffed endotracheal tube. No lesions were encountered during bronchoscopy of the vocal cords or elsewhere. The patient was then induced with Inj. Propofol 110 mg IV, Inj. Fentanyl 100 mcg IV, and Inj. Cisatracurium 8 mg IV. Anaesthesia was maintained with sevoflurane 1%, nitrous oxide at 1 L/min, and oxygen at 1 L/min.

Postprocedure, we ensured that the patient had adequate spontaneous breathing efforts, after which we reversed her neuromuscular blockade with Inj. Neostigmine 2.5 mg IV and Inj. Glycopyrrolate 0.5 mg IV. Following adequate reversal and suctioning, we extubated the patient. Postoperatively, she was fine, had no complaints, and was discharged on postoperative day 5.

DISCUSSION

Calcinosis Cutis is the deposition of calcium salts in the skin and subcutaneous tissues, first described by Teissier in 1877 [1]. When it is distributed widely throughout the body, it is known as Calcinosis Cutis universalis and is encountered more frequently in females than in males, especially in younger age groups [2]. It has various types, including idiopathic, iatrogenic, metastatic, and dystrophic [3]. The possible mechanisms for calcium deposition may include hypoxia, repeated trauma, cellular damage, and acidic pH leading to suppression of calcification inhibitor molecules [4]. Calcinosis Cutis is commonly associated with autoimmune disorders, particularly dermatomyositis and systemic sclerosis [2], and, though rarely, with systemic lupus erythematosus [5]. Literature findings are compared in [Table/Fig-2] [6-9].

Authors	Findings	Comparisons
Khan QA et al., [6]	The author reported a case of juvenile dermatomyositis with muscular weakness at 3 years of age, following which the patient developed Calcinosis Cutis at the age of 8.	In this case, the patient was first diagnosed with dermatomyositis at 7 years of age and later diagnosed with Calcinosis Cutis at 11 years of age.
Nanjundaswamy NH et al., [7]	The author encountered a difficult airway in a dysmorphic Calcinosis Cutis of the gluteal region without any association with connective tissue disorders, posted for surgical excision and flap cover. Unlike our case, which was a clear-cut case of difficult airway, theirs showed no degree of difficulty during airway examination. Hence, they tried intubation with a laryngoscope, which failed, and during the second attempt, they intubated with a MacCoy blade 3.	In this case, we immediately planned awake fiberoptic intubation due to the difficulty.
Wong AV et al., [8]	Author reported a pregnant female with Calcinosis Cutis in active labour for labour analgesia. They gave an epidural labour analgesia for pain relief. In this case, since it was a nasal bone fracture reduction, we could not do regional anaesthesia.	In this case, since it was a nasal bone fracture reduction, we could not do regional anaesthesia.
Lagoo JY et al., [9]	Author reported a case of Calcinosis Cutis universalis on the right arm posted for surgical excision of the lesion. They did an ultrasound-guided right supraclavicular block since the patient had restricted mouth opening and neck mobility.	Since the patient was posted for nasal bone fracture reduction, general anaesthesia was done.

[Table/Fig-2]: Comparison of anaesthetic challenges and techniques in reported cases of Calcinosis Cutis [6-9].

at 11 years of age. Similarly, Khan QA et al., reported a case of juvenile dermatomyositis with muscular weakness at three years of age, followed by the development of Calcinosis Cutis at the age of eight [6].

Nanjundaswamy NH et al., encountered a difficult airway in a case of dysmorphic Calcinosis Cutis in the gluteal region, without any association with connective tissue disorders, who was posted for surgical excision and flap coverage [7]. Unlike our case, which presented a clearcut instance of a difficult airway, theirs showed no difficulties during airway examination. They attempted intubation with a laryngoscope, which failed, and during the second attempt, they successfully intubated using a McCoy blade 3. In our case, we immediately planned for awake fiberoptic intubation due to the difficulties presented.

Wong AV et al., reported a pregnant female with Calcinosis Cutis in active labor for analgesia [8]. They administered epidural labor analgesia for pain relief. In our case, since it involved nasal bone fracture reduction, we could not perform regional anaesthesia. Similarly, Lagoo JY et al., reported a case of Calcinosis Cutis universalis on the right arm, who was posted for surgical excision of the lesion [9]. They performed an ultrasound-guided right supraclavicular block because the patient had restricted mouth opening and neck mobility, unlike in this case.

According to our literature search, there are very few reported cases of Calcinosis Cutis undergoing surgery [7,9]. In connective tissue disorders, an anaesthesiologist may face numerous challenges, such as difficult airway management due to restricted mouth opening and neck movement from collagen deposition and fibrosis. Additionally, stiffness of joints can complicate positioning during surgery, and difficulty in obtaining venous access may arise due to lesions. Systemic sclerosis can be associated with CREST syndrome, requiring extra care to avoid aspiration and mucosal bleeding during intubation [9,10]. This disease currently cannot be cured, and effective strategies or therapies are still being sought.

The treatment options for Calcinosis Cutis include both medical and surgical therapies. The first line of medical management typically involves calcium channel blockers like diltiazem, colchicine, minocycline, bisphosphonates, warfarin, and intravenous immunoglobulins. Surgical excision may also be beneficial, although the rate of recurrence is high. Ulcerated lesions are treated with wet acetic acid dressings and systemic antibiotics for any active infections. Physical therapy may provide pain relief and enhance joint mobility [4].

CONCLUSION(S)

Calcinosis Cutis universalis is a rare clinical phenomenon, and patients requiring anaesthesia for surgeries are even rarer. These patients may present with a difficult airway and challenges for regional anaesthesia due to extensive lesions. Therefore, careful evaluation is essential to identify potential airway difficulties or challenges with regional anaesthesia to avoid anaesthesia-related mortality and morbidity. This case report highlights how connective tissue disorders can pose significant challenges for anaesthesiologists, but such challenges can be effectively managed when the physician is fully prepared.

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