



Tibial shaft fracture in rare case of Congenital Methemoglobinemia mimicking covid like illness , and its effect on Bone Healing : A Case Report

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Abstract: Methemoglobinemia is an uncommon, often overlooked, and potentially reversible cause of hypoxia in the perioperative setting. Methemoglobinemia occurs when the bound ferrous iron (Fe²⁺) of oxyhemoglobin is oxidized to the bound ferric iron (Fe³⁺) of methemoglobin. An indication of methemoglobinemia is a discrepancy between the transcutaneous oxygen saturation (SpO₂) and the calculated arterial oxygen saturation on arterial blood gas (SaO₂). Patients with lifelong congenital methemoglobinemia or with a history of chronic methemoglobinemia secondary to chronic exposure to drugs or toxins, methemoglobin levels can be as high as 40% and still be well tolerated with cyanosis (blueish cast to the mucous membranes of the skin) being the only presenting manifestation. No reports with regards to the effect of methemoglobin during pre-operative, intra-operative or post-operative period have been described and the effect of bone healing in methemoglobin patient have not been described in literature. It mimics covid like illness with decreased saturation, at presentation resulting in being not fit for surgery however after thorough investigation it was diagnosed to be methemoglobinemia.

Here we present a case of methemoglobinemia mimicking covid illness with tibia and fibula fracture with 2 years follow-up with no intra-op, post-op complication which later went for union of fracture 2 years post primary surgery.

Introduction: Methemoglobinemia is an uncommon, often overlooked, and potentially reversible cause of hypoxia in the perioperative setting. Methemoglobinemia occurs when the bound ferrous iron (Fe²⁺) of oxyhemoglobin is oxidized to the bound ferric iron (Fe³⁺) of methemoglobin⁽¹⁾⁽²⁾⁽³⁾⁽⁴⁾. An indication of methemoglobinemia is a discrepancy between the transcutaneous oxygen saturation (SpO₂) and the calculated arterial oxygen saturation on arterial blood gas (SaO₂)⁽¹⁾. The diagnosis may be complicated by the effect of MHb on arterial blood gas and pulse oximeter oxygen saturation results⁽⁵⁾⁽³⁾. Patients with lifelong congenital methemoglobinemia or with a history of chronic methemoglobinemia secondary to chronic exposure to drugs or toxins, methemoglobin levels can be as high as 40% and still be well tolerated with cyanosis (blueish cast to the mucous membranes of the skin) being the only presenting manifestation⁽¹⁾⁽⁶⁾. Methemoglobin doesn't bind oxygen and hence cannot deliver oxygen to tissues. Patient with methemoglobinemia is asymptomatic except for cyanosis. It is suspected in the presence of dyspnea or unexplained cyanosis and hypoxemia that does not improve with supplemental oxygen. The severity of the presentation may vary from asymptomatic cyanosis in mild cases to severe distress⁽⁷⁾⁽⁸⁾. Any patient with total Hb of 10 g/dL with 10% methemoglobin will have total methemoglobin of 1 g/dL and will not be cyanotic. Tibial shaft fractures account for 36.7% of fractures of long bones and over 2% of all fractures. Intramedullary nailing of the tibia is the standard of care for tibial fractures as it ensures rapid bone healing and an expedited recovery⁽⁹⁾⁽¹⁰⁾. No reports with regards to effect of methemoglobin during pre-operative, intra-operative or post-operative period have been described and the effect of bone healing in methemoglobin patient have not been described in literature Here we present a case of methemoglobin mimicking covid illness with tibia and fibula fracture with 2 years follow-up with no intra-op, post-op complication which later went for union fracture 2 years post primary surgery during both primary surgery and during implant removal.

Case Report

A 41 years old male complains of pain and swelling in right leg following RTA. Pain was sudden in onset, gradually progressive, aggravated with movement and relieved with rest and medication. He is a known case of T2DM. On general examination his saturation was found to be 55% at RA with no cyanosis, on local examination gross deformity of right leg was noted with no wound, swelling with crepitus present and abnormal mobility present in right leg (distal 1/3rd). On hematological investigation blood parameters were found to be normal and on X-Ray of right leg fracture of right tibia and fibula at junction of mid and distal 1/3rd was planned for surgery but pulmonology and cardiology opinion was taken i/v/o low saturation was taken and relevant investigation was done and COVID test was negative and 2D echo was normal with ejection fraction of 60%. After proper investigation and ruling out covid and other condition related to low saturation and confirming methemoglobinemia, patient was taken-up for surgery after anesthetic clearance. He was operated with closed reduction and internal fixation with IMIL nail with preparation to tackle any complication that could have happened during surgery. There was no intra-op, post-op complication and saturation in post-op holding was 60% at room air and he was shifted to the ward after 4 hours. He was discharged after 2 days post-surgery and was on regular follow-up with no early or late complication and staples were removed after 12 days of surgery and was on monthly follow-up till 3 months post-op during which radiographs showed evidence fracture healing with callus. He returned to work after 4 months post-surgery. He

visited to us again after 2 years after a blunt trauma to right leg with swelling noted over right leg and X-Ray of right leg was taken and showed united tibia and fibula fracture with IMIL in-situ (Figure 1) and his blood parameters like Hb- 12.0 g/dL, platelets- 2.24 lakh/mm³, HbA1c- 13.0 other all parameters were within normal limits with saturation of 55% at room air. After obtaining clearance from Pulmonologist and Anesthesiologist he was planned for implant removal and taken for implant removal with no intra-op and post-op complication. Figure 3 shows recent intra-operative monitoring of the patient with saturation of 55% with oxygen via mask. Post-operative period was uneventful with no signs or symptoms of dyspnea and patient was immediately shifted to post-operative holding for further management and was shifted to the wards after 4 hours. Post-operative X-ray showed now evidence of fracture with evidence of implant removal. He was on regular follow-up post-surgery and showed no signs of surgical site infection and sutures were removed 14 days after surgery and was started with partial weight bearing mobilization from 2nd day and full weight bearing after suture removal. He returned to his work 3 weeks after surgery.



Figure 1 : Plain X ray showing tibia and fibula fractures



Figure 2: Plain X ray showing union and healed fractures of tibia and fibula after closed reduction and internal fixation

DISCUSSION: Methemoglobinemia can be either congenital or acquired. The most common cause of congenital methemoglobinemia is a result of deficient cytochrome b5 reductase (CYB5R). CYB5R deficiency is a result of variations in the cytochrome b5 reductase 3 gene (CYB5R3). The variations lead to type 1 and type 2 CYB5R3. Type 1 focuses on the CYB5R deficiency in RBCs. Patients are typically asymptomatic aside from appearing cyanotic⁽⁷⁾. Some patients with congenital methemoglobinemia will develop physiological adaptations and as result will be asymptomatic even with an elevated level of methemoglobin (up to 40%) due to changes in the concentration of 2,3-Diphosphoglycerate and pH, synthesis of globin chains and polycythemia⁽³⁾. Congenital methemoglobinemia is usually missed clinically. Oftentimes congenital methemoglobinemia presents in infancy in contrast to acquired methemoglobinemia which presents in older age groups⁽⁷⁾. In our case the patient was reported to be diagnosed with methemoglobinemia during pre-surgery check-up 2 years back after the road traffic accident during COVID endemic with low saturation which mimicked COVID like illness but on further evaluation it was found to be congenital asymptomatic methemoglobinemia.

Tibial shaft fractures account for 36.7% of fractures of long bones and over 2% of all fractures⁽⁹⁾. As it is a common fracture affecting all the age groups and mostly male population⁽¹⁰⁾ and methemoglobinemia being a rare and underreported illness , knowing about the effect of methemoglobinemia on complications encountered during surgery and factors affecting fracture healing can lead to better treatment to the patient as in our case there was no complications reported during primary surgery after the fracture and even during second surgery during implant removal. Hence, methemoglobinemia patient can safely be treated with surgical management without any complication either during intra-operative/post-operative period.

CONCLUSION:

Fracture healing with operative management in a methemoglobinemia patient does not have any intra-op and post-op complication and union was also achieved at fracture site at 2 years follow-up.

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