

# Infected Open Depressed Skull Fracture Complicated with Tetanus Grade I in an Unimmunized Child: A Rare Case Report with Literature Review

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## Case report

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# Abstract

**Background:** Tetanus is a rare disease caused by *Clostridium tetani*, which produces tetanolysin and tetanospasmin. In 2018, there were only approximately ten tetanus cases reported in Indonesia. Despite widespread vaccination, especially in low-middle income countries, tetanus still occurs (mostly in adult) due to the lack of immunization related to religious tenets, cultural belief or inaccessibility to medical care. In addition, tetanus in pediatric population shows features which are quite distinct from the adult group.

**Case Description:** We reported a 7 year old girl that presented to our institution with a history of falling 10 days prior the admission, with only skin laceration of her forehead. Since one day prior to admission, the patient looked drowsy and difficult to be awakened, accompanied with stiffness of her jaw; we diagnosed as unimmunized child with open depressed skull fracture of her frontal bone and wound infection complicated with "lockjaw". Perioperative management of that rare case is reported and discussed.

**Conclusion:** This case is so exceptional, since the infected open depressed skull fracture complicated with trismus in an unimmunized child occurrence in our center is extremely rare; the pediatric intensive care of such patients requires halting further toxin production, neutralisation of circulating toxin and control of the clinical manifestation induced by the toxin that has already gained access to the central nervous system. The basic tents of anaesthetic care in such case had to well managed and planned prior the surgery.

## Introduction

Tetanus is an infection that involved the nervous system, caused by a bacterium known as *Clostridium tetani* (*C. tetani*). It is different from other vaccine-preventable diseases because it does not spread from person to person. Spores of tetanus bacteria are everywhere in the environment and enter into the body through damage of skin. The spores develop to become bacteria when it enters the body usually cuts or puncture wounds caused by contaminated objects. Tetanolysin damages surrounding tissue and is also capable of haemolysis. Although tetanolysin plays no direct role in the clinical manifestations of tetanus, it is thought to optimise milieu for bacterial proliferation. Tetanospasmin (metalloprotease) produced by this bacterium travels through axons in a retrograde fashion to CNS leads to unopposed muscle contraction and spasm. Seizures may occur and the autonomic nervous system may also be affected. This is a preventable disease with available vaccinations. Nearly all tetanus cases are among people who did not get the recommended tetanus vaccination; include people who have never received a tetanus vaccine and adults who do not have their 10-year booster shots. Although prevention been incorporated in our national immunization program by Indonesia Health Ministry, tetanus remains one of our major health problem. World Health Organization reported that there were 391 cases of tetanus in children and neonates in Indonesia in 2019.<sup>1,2</sup> In this study, we reported a case report of a seven years-old girl with inadequate immunization status that had a head injury that initially received suboptimal wound care, which led to tetanus and performed craniotomy debridement. Herein, we discussed the clinical presentation and management of the patient according to the literature.

## Case Report

A seven-years-old girl was admitted to our hospital, Dr. Hasan Sadikin Hospital, with decreased consciousness. Ten days prior to admission, when the patient was playing at a public restroom in Majalaya, she lost her balance and fell over with her head hit the floor that was made of cement. She had a skin laceration at her forehead, she was brought to a nearby primary healthcare facility, underwent wound suturing, and discharge with uneventful symptoms. Five days afterwards, she had a fever and purulent discharge from her wound, she was brought to the local hospital, underwent wound cleansing, and then discharged home. One day prior to admission, the patient looked drowsy and difficult to be awakened, accompanied with stiffness of her jaw, she was brought to another hospital. She received anti seizure medication intravenously and then referred to our hospital.

The patient had never received any vaccination since birth because the midwife was uncertain for conducting vaccination to low-birth-body-weight babes. Another issue, due to some religious view, her family rejected any vaccination for her. At the admission, her vital sign was within normal limits. There was a purulent wound at her left frontal with  $2 \times 1.5 \times 0.5$  cm dimension and on palpation there was bone discontinuity; from neurological examination, she exhibited nuchal rigidity, trismus  $> 1$  finger and motoric spasticity. Other tetanus signs such as spontaneous spasm, provoked spasm, board-like abdomen, and opisthotonus were absent. There was no autonomic dysfunction; laboratory results was mainly uneventful, except thrombocytosis ( $718 \times 10^9$  cells/L); skull x-ray shown a double counter appearance at her left frontal (Fig. 1); by contrast to head CT scan, a depressed skull fracture  $> 1$  table was confirmed without any enhancement (Fig. 2).

The patient was diagnosed as open depressed skull fracture and tetanus grade I. For the tetanus treatment, the patient was given Human tetanus Immunoglobulin (HTIG) 3000 IU, Ampicillin Sulbactam  $4 \times 1$  g IV, Metronidazole  $3 \times 200$  mg IV and diazepam  $4 \times 2$  mg IV. For depressed skull fracture, she underwent craniectomy debridement. The patient underwent general anaesthesia and endotracheal intubation; as anesthesiologist prepared fiberoptic bronchoscopic, meanwhile otolaryngologist was ready for tracheostomy in case the intubation attempt was failed. Fortunately, endotracheal intubation was successfully performed with a 5ID cuffed PVC endotracheal tube. The patient was premedicated with  $60 \mu\text{g}$  of fentanyl, while the induction was performed with 50 mg of propofol and 20 mg of rocuronium. Throughout the operation, the patient was ventilated with  $\text{O}_2$  plus  $\text{N}_2\text{O}$  (50:50) and 2% sevoflurane. The "lockjaw" trismus made intubating attempt to the patient challenging, hence an extra dose of rocuronium (1 mg/kgBW) was given.

Intraoperatively, we found a fragmented interlocking fracture, with a dimension of  $3 \times 2$  cm. Then, craniectomy was performed with boundaries of healthy bone, the duramater was yellowish, intact, and not tense. Bone defect was to no reconstruct and proceeds with skin closure (Fig. 3). Pus were then sampled from the fracture location and sent for staining examination and culture. However, both examinations showed no microorganism growth. Postoperatively, the patient sent to pediatric intensive care unit for 3 days with mechanical ventilation and then transferred to ward. After 3 weeks post-

operative patient showed remarkable clinical recovery. After discharge from hospital, the patient is suggested to get the tetanus toxoid vaccine in primary health care.

## Discussion

Tetanus is an acute disease affecting the CNS and is now a rare disease in the western world, but not so in developing countries. Each year, approximately 2.8 million people sustain head injuries in the United States alone, resulting in approximately 2.5 million emergency evaluations, 300,000 hospital admissions and 60,000 deaths. There are several factor in our society that play role in high incidence of tetanus, there is still some religious tenets, cultural belief, and traditional healer that act as barrier to immunization and good wound care barrier, as to mention although in this case these factors did not contribute, and the barriers to immunization in this case were midwives uncertainty for conducting vaccination to low-birth-body-weight baby, and mother in this case is passively accept with resignation that her children didnt received immunization due to midwives uncertainty. This is the first reported case tetanus complicating an untreated mild open head injury in Indonesia that underwent craniotomy debridement for source control .<sup>3</sup>

Tetanus primary manifestation is prolonged muscular spasm (without altered mental status) caused a neurotoxin produced by *C. tetani* that potentially given fatal complication. *C. tetani*, a Gram-positive anaerobic bacterium, exists as sporulated form in environment (usually in soil) throughout the world and found in gastrointestinal tract of animals and human. *C. tetani* spores enter the body through a wound or damage skin; in the presence of anaerobic conditions, the spores germinate. The bacteria produce two very potent toxins (tetanolysin and tetanospasmin), both toxins enter the blood stream and lymphatic system to disseminate through the body. Tetanolysin is thought to optimize conditions for bacterial proliferation. The clinical features of tetanus are caused by tetanospasmin, enters the peripheral nervous system directly from the contaminated wound, and is capable of affecting motor, sensory, and autonomic neurons. In approximately 20% of cases, no entry wound is noted. The primary pathological effect of tetanospasmin is the cleavage of synaptobrevin, which is a presynaptic protein. Synaptobrevin facilitates neurotransmitter fusion of vesicles to nerve membranes and release of their contents into the synapse. By cleaving synaptobrevin, neurotransmission is effectively blocked. Radiolabelled assays have shown tetanospasmin has a preference for inhibitory motor neurons, which explains the clinical picture of muscle rigidity. Toxins act at several sites within CNS, including peripheral motor end plates, spinal cord, and brain, as well as in the sympathetic nervous system. Toxin causes tetanus typical clinical manifestations by interfering with the release of neurotransmitters and block inhibitor impulses; leads to unopposed muscle contraction and spasm. Seizures may occur and the autonomic nervous system also is affected.<sup>1,4,5</sup>

After inoculation the incubation period ranges from 3 to 21 days, averaging about 10 days. In general, the further the injury site is from the central nervous system, the longer the incubation period. A shorter

incubation period is associated with more severe disease complications and a higher mortality. In neonatal tetanus, symptoms usually appear from 4 to 14 days after birth, averaging about 7 days. The characteristic symptoms of tetanus are painful muscular contractions, primarily of the masseter and neck muscles and secondarily of trunk muscles. Trismus or “lockjaw”, is a common sign of tetanus. A common first sign suggestive of tetanus in older children and adults is abdominal rigidity, although rigidity is sometimes confined to the region of injury. Generalized spasms occur frequently induced by sensory stimuli. History of an injury or apparent port of entry may be lacking. Clinicians rarely recover the organism from the site of infection.<sup>1,4,5</sup>

There are four clinical features of tetanus: neonatal, generalized, localized, and cephalic; four Grade of tetanus as propose in Ablett Classification of tetanus severity: I (mild), II (moderate), III (severe), and IV (very severe) as we can see in Table 1. **Neonatal tetanus** occurs 3–7 days postdelivery. Difficulty feeding, poor suck/swallow, excessive crying often precede overt spasms. **Generalized tetanus** is the most common feature, accounting for > 80% of cases. The most common initial sign is spasm of the muscles of the jaw or “lockjaw”. Other signs may follow “lockjaw” from such painful spasms in other muscle groups in the neck, trunk, extremities, and generalized, seizure-like activity or convulsions in severe cases. Even with modern intensive care, generalized tetanus is associated with death rates of 10–20%. **Localized tetanus** is an unusual feature with consisting of muscle spasms in a confined area close to the site of the injury. Although localized tetanus often occurs in people with partial immunity and is usually mild, progression to generalized tetanus can occur. **Cephalic tetanus is the rarest feature** associated with lesions of the head or face and may also be associated with otitis media. The incubation period is short, 1–2 days. Unlike other, cephalic tetanus results in flaccid cranial nerve palsies rather than spasm, but spasm of the jaw muscles may also be present. Like localized tetanus, cephalic tetanus can progress to the generalized form. most cases of cephalic tetanus involve only the facial region below the eyebrows in CN V (trigeminal nerve) territory, in the periorbital region. Few cases attributable to the head injury usually were secondary to small scalp lacerations of the frontal temporal regions as a result of minor trauma. Cases secondary to open depressed skull fracture, such as our case, are almost unknown.<sup>5-17</sup> Study from ethiopia showed us that trauma is the most common portal of entry in children as much as 79.2%. This can be explained by the high chance of ignoring pediatric trauma and lack of provision of tetanus prophylaxis. The most common cause of death was the respiratory failure secondary to uncontrolled spasm (laryngo- and diaphragm spasm); could be due to muscle relaxant poor escalation and lack of pediatric intensive care unit (ICU) care.<sup>1,18-20</sup>

Tetanus disease never is completely eradicated because the bacteria maintain natural reservoirs in soil, humans, and other animals. Furthermore, there is no naturally acquired immunity; the very small amount of toxin necessary to cause disease does not stimulate antibody production in the host. Tetanus is almost entirely vaccine preventable since the vaccine efficacy is virtually 100%. A 6-dose series of tetanus toxoid-containing vaccine (TTCV)-which includes 3-dose in the first year after birth, followed by booster dose in the 2nd year, in early childhood, and in adolescence has been shown to provide immunity and protect women through their childbearing years, though some countries still endorse a 5-dose series

(shown in Fig. 4). Indonesia national immunization programs include 5-dose TT vaccination. Three-dose given before the first year after birth, boosted at 4–7 and 15 years old. Vaccination and good wound care are the main feature to prevent tetanus.<sup>4,21</sup>

The treatment of tetanus involves providing supportive care for symptoms of muscle spasms and potential respiratory compromise, neutralizing the remaining tetanus toxin, and good wound care for eradicating bacteria at the wound site. The defining factor that contribute to therapeutic success was time for immunoglobulin administration to neutralize the toxin and/or tetanus toxoid vaccination as we can see in table 2. Without treatment, the case fatality rate remains ~ 100%; with treatment, case fatality rates drop to 10–20%. For generalized tetanus case, intensive care, including endotracheal intubation, mechanical ventilation, deep sedation and/or paralysis is the mainstay of supportive care. Patients should have minimal environmental stimulation to avoid the reflex spasms. Benzodiazepines, particularly diazepam, are treatments of choice, since provide sedation, control muscle spasm, and provide anxiolysis. Intravenous magnesium sulfate can be used as an adjunct anti spasmodic agent that also decreases autonomic instability. Intrathecal baclofen has shown promise for severe spasms, but may not be feasible in resource-poor areas where intrathecal catheter insertion isn't practical and mechanical ventilator support is unavailable; higher dose cause respiratory depression and cardiovascular instability. Autonomic instability can be treated with clonidine, b-blockers, and morphine.<sup>4,22-23</sup>

Rapid sequence intubation is mandated due to significant aspiration risk because of increased abdominal pressure, gastric stasis, and involvement of laryngeal muscles in tetanus patient. Preparation for a potentially difficult airway was in place and spontaneous ventilation was maintained by the induction of anaesthesia with sevoflurane in 100% O<sub>2</sub>. When general anaesthesia is performed, a deep level of anaesthesia is suggested to avoid triggering hypertensive crises and spasms during the procedure. Various intravenous and inhalation anaesthetic agents have been used without incidence in tetanus patients.<sup>24</sup> In our case, there are several risk factor related to tetanus complications. Firstly, she never received any vaccination and the wound is not properly taken care of, moreover she did not get immunoglobulin to neutralize the toxin after the accident. All the factors above contribute to tetanus; the patient underwent craniectomy, debridement for source control under neuroanesthesia and post-operative care in pediatric ICU.

The pediatric intensive care of such cases requires halting further toxin production, neutralisation of circulating toxin, and control of the clinical manifestation induced by the toxin that has already gained access to the CNS. The basic tenets of anaesthetic care in a patient with tetanus include consideration of the implication of a full stomach, prevention of the paroxysm of muscle spasm by maintaining a deep plane of anaesthesia or the use of regional anaesthesia and control of the autonomic instability. As the autonomic instability may be life-threatening, invasive arterial blood pressure monitoring may be indicated. Although uncommon, with prolonged tetanus, associated cardiomyopathy repeated exposure to catecholamines may be present. A perioperative assessment of renal and electrolyte balance required the presence of myoglobinuria and renal dysfunction. In our case, three weeks postoperative course was

uneventful and the patient showed an excellent clinical recovery and discharged on 16th postoperative day.

## Conclusion

In summary, we present a rare case of a seven years old unimmunised girl with open depressed skull, fracture of frontal bone, and wound infection complicated with “lockjaw”. This case report highlights the crucial role that proper, optimal wound management, and tetanus immunization in children have in preventing tetanus. Furthermore, this report also points out the need to improve the public awareness and knowledge of hygiene, sanitation, and the monumental benefits of immunization on ones child’s life.

## Abbreviations

CNS: central nervous system; CT: computed tomography; PVC: polyvinyl chloride; CN: central nervous; TT: tetanus toxoid

## Declarations

## Acknowledgements

None

## Authors’ contributions

Corresponding author: Dzulfikar DLH was one of the two prehospital physicians on the incident and is the primary author of this manuscript. DLH finished the manuscript for publication. All authors treated the patient. EHL, AF, IM and MZA performed the surgery. All authors participated in writing the manuscript, have read and approved the final manuscript.

## Authors’ informations

DLH is a consultant of pediatric emergency and intensive care in Hasan Sadikin General Hospital. He is the head of pediatrics residency programme in Padjajaran Univeristy/Hasan Sadikin General Hospital. AF is a neurosurgeon in Hasan Sadikin General Hospital. He has a special interest for neurotrauma neurosurgery. AN is a pediatric residency in Hasan Sadikin General Hospital. EHL is neurosurgery residency in Hasan Sadikin General Hospital. MZA is a neurosurgeon in Hasan Sadikin General Hospital. He is the head of Neurotrauma Division Neurosurgery Departement. AI is a neurosurgeon in Hasan Sadikin General Hospital. He has a special interest for infectious neurosurgery. IA is an anesthesiology in Hasan Sadikin General Hospital. He has a special interest for neurotrauma neurosurgery for neuroanesthesi. He is the head of educational research unit of Hasan Sadikin General Hospital.

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## Availability of data and materials

Not applicable

## Ethics approval and consent to participate

Informed consent was obtained from the patient parents for publication of this case report and any accompanying images. Her family was present at the time

## Consent for publication

We have an attachment from the parents that consent this case report for publication.

## Competing Interests

The authors declare that they have no competing interests.

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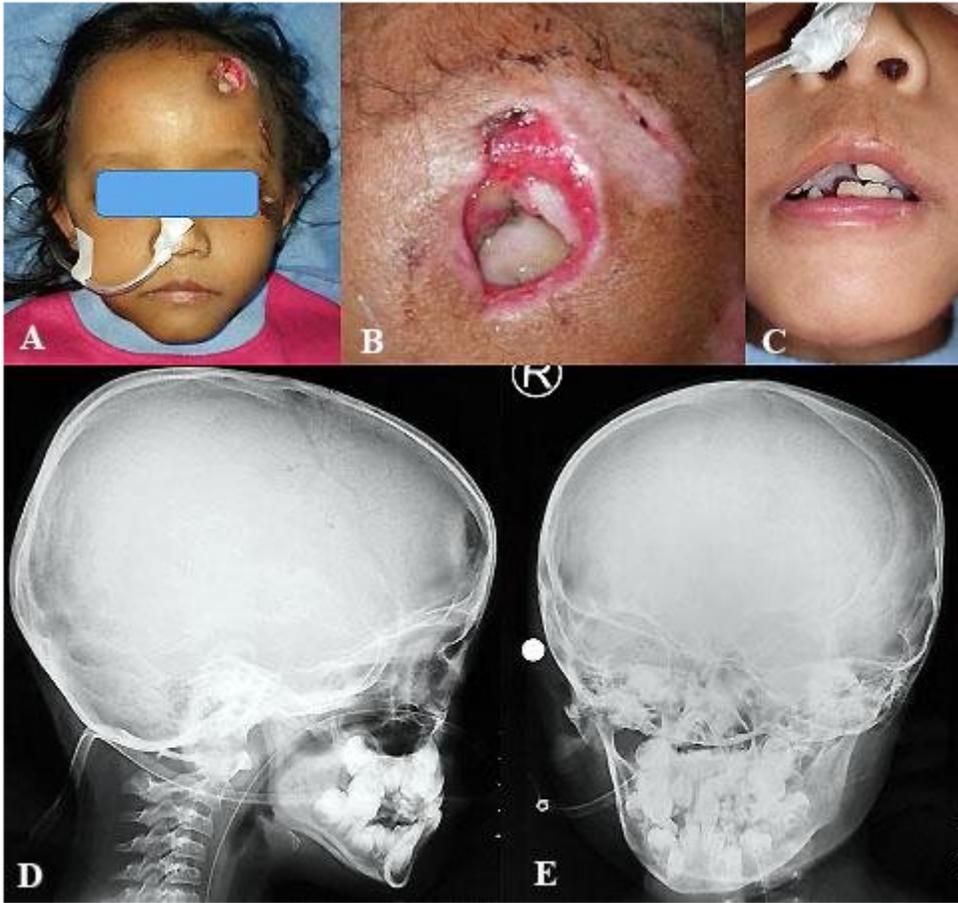
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## Figures



**Figure 1**

Representative images of patient condition. A) There was a purulent wound at her left forehead. B) Higher magnification, showed the condition of her wound. C) She had a trismus with less than 1.5 cm breadth. D-E) Skull X-Ray at Hasan Sadikin Hospital showed a double contour appearance at left frontal



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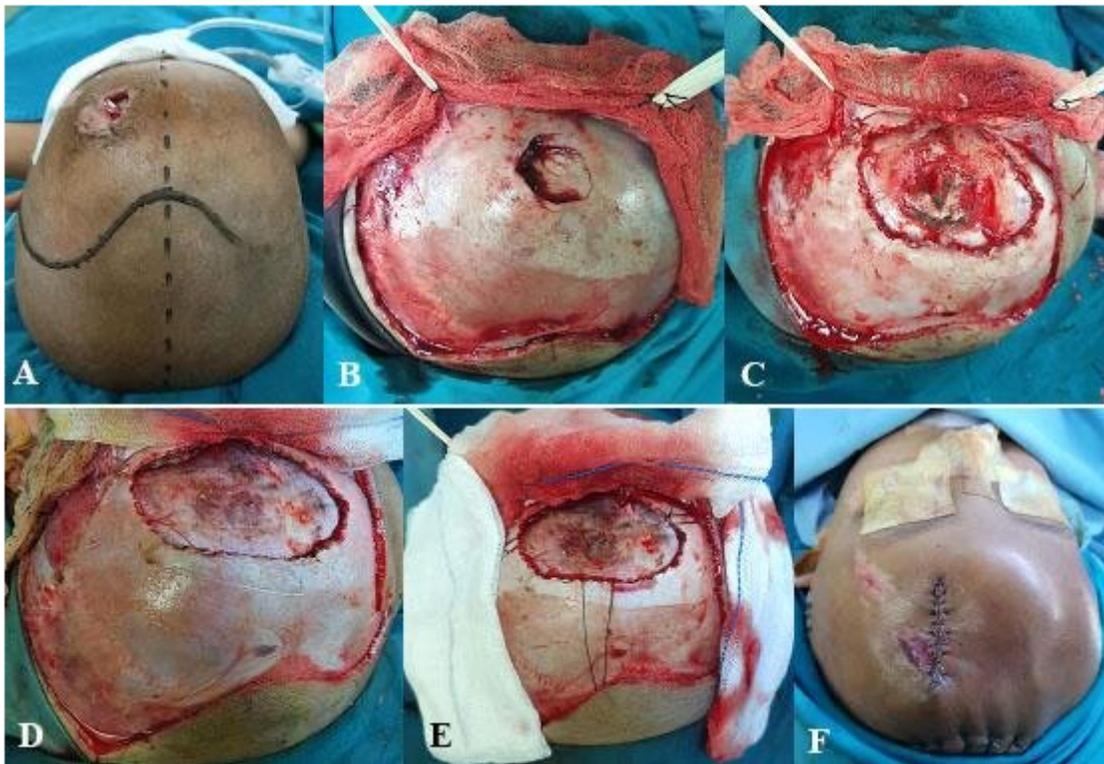
**Figure 2**

Contrast Head CT Scan showed a depressed skull fracture of left frontal bone.



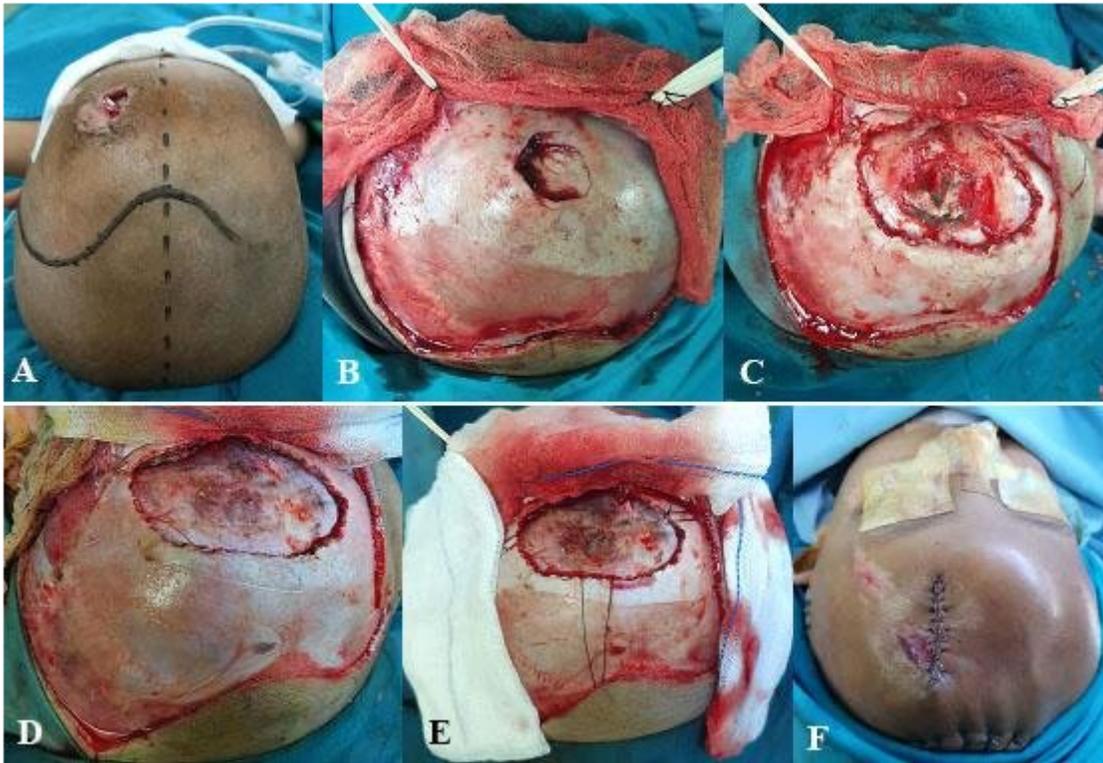
**Figure 2**

Contrast Head CT Scan showed a depressed skull fracture of left frontal bone.



**Figure 3**

A) The patient was positioned supine and ¾ coronal incision was performed. B) A depressed interlocking skull fracture about 3x2 cm. C) Craniectomy was performed, the duramater was yellowish, intact, and not tense. D) Debridement was conducted. E) Bone defect was not repaired. F) The wound and incisional wound was primarily sutured.



**Figure 3**

A) The patient was positioned supine and  $\frac{3}{4}$  coronal incision was performed. B) A depressed interlocking skull fracture about 3x2 cm. C) Craniectomy was performed, the duramater was yellowish, intact, and not tense. D) Debridement was conducted. E) Bone defect was not repaired. F) The wound and incisional wound was primarily sutured.

# People of all ages need TETANUS VACCINES



**DTaP**  
for young children

- ✓ 2, 4, and 6 months
- ✓ 15 through 18 months
- ✓ 4 through 6 years

**Tdap**  
for preteens

- ✓ 11 through 12 years

**Td**  
for adults

- ✓ Every 10 years

[www.cdc.gov/tetanus](http://www.cdc.gov/tetanus)



Figure 4

This graphic image icon highlights CDC's tetanus vaccination recommendations for young children, preteens, and adults.

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