International Journal of Surgery Case Reports 88 (2021) 106484

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.elsevier.com/locate/ijscr

Case report

Jael's syndrome: Case report and literature review

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ARTICLE INFO

Keywords:
Jael's syndrome
Retained knife
Assault
Frontal region

ABSTRACT

Introduction and importance: Jael's syndrome, an intentional injury caused by a knife in the face or skull is a rare encounter in clinical and forensic practice, rarely involving an impacted knife. Clinical and radiographic diagnosis is essential to identify severity of injury and location of the retained knife. To the best of our knowledge, this is the first reported novel case of Jael's syndrome in Tanzania.

Case presentation: We present the case of a 31-year old man admitted at Muhimbili National Hospital following an impacted knife. The stab wound extended to the medial wall of left orbit and ended just before the optic foramen associated with vitreous hemorrhage and the retained knife caused superoposterior displacement of the globe. Multidisciplinary management was instituted including prompt evaluation, imaging and surgical removal of the knife under general anesthesia.

Clinical discussion: Plain skull X-ray revealed an extensive retained blade and computerized tomography (CT) showed the tip of the blade adjacent to the right styloid process with no neurovascular compromise. Initial concern was the left eye that was reported to be viable by ophthalmologists. Incredibly, the patient had no initial sequelae from such an extensive injury and had unremarkable recovery with no complications apart from the wound to left inferior rectus muscle that was conservatively managed. Simple withdrawal of the retained knife was successful.

Conclusion: Craniofacial retained knives are rare. Thorough prompt initial evaluation and intervention is vital since improper management can be devastating.

1. Introduction

Jael's syndrome was described by Jefferson in 1968 as an intentional injury caused by a knife in the skull or face [1,2]. Penetrating knife injuries to the face are rarely reported in many parts of the world including Tanzania. A retained knife following a stab wound to the face remains to be an uncommon injury in clinical and forensic practice and also challenging to the clinician when associated with significant vascular and neurological dysfunction [1–6].

This term "Jael's syndrome" has been related to the bible passage about the homicide of Sisera by Jael described in Judges IV:21 (Jerusalem bible, Portuguese edition) [1,2].

Even though rare, stab wounds in the face due to retained knives are often sequelae of assaults or occupational accidents. Depending on the extent of such penetrating injury, the blade may cause critical damage to vascular structures and therefore imaging studies are fundamental to evaluate the extent of the injury such as localization of the blade and whether it has cause damage to nearby vital structures [1,3,7–10].

A thorough initial assessment coupled with prompt intervention is of paramount importance since inappropriate management can result in complications such as diplopia, blindness, intracranial hemorrhage and acute cerebrospinal fluid (CSF) leakage [1,8,9,11,12]. Imaging with two plain films in different planes can help to localize the knife and the indications for subsequent imaging with CT include low density foreign bodies and deep penetrating injury with metallic foreign bodies [1,2] as exemplified by this case report.

Multidisciplinary management has to be instituted when such retained foreign bodies are encountered and this has to include a stable airway, assessing hemodynamic stability of the patient and the extent to which vascular and neuro-ophthalmological structures have been

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https://doi.org/10.1016/j.ijscr.2021.106484
Received 6 September 2021; Received in revised form 30 September 2021; Accepted 3 October 2021
Available online 7 October 2021
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compromised. The work has been reported in line with the SCARE 2020 criteria [13].

2. Case presentation

A 31-year old man presented at the Emergency Department of a National Hospital 16 h following an assault. He was stabbed at the frontal aspect of the face and presented with severe headache and bleeding from the injured site. Upon arrival, he had hemodynamic instability due to acute blood loss though his airway was stable. Initially, plain skull X-rays were ordered and revealed an extensive retained knife. Consequently, CT scans of the head revealed a penetrating injury to the left frontal bone, extending to the medial wall of the left orbit and ending just before the optic foramen associated with vitreous hemorrhage. The margin of the retained knife blade caused superoposterior displacement of the globe and the tip of the blade had reached the right styloid process with no neurovascular compromise.

His medical history was remarkable for severe headache and bleeding from the injured site and physical examination revealed no abnormalities in the respiratory system.

Results of laboratory tests showed anemia due to acute blood loss (hemoglobin, 7 g/dl), HIV serology was negative. Serum creatinine, blood urea nitrogen and electrolytes were within normal ranges.

Immediate management of the patient was instituted through multidisciplinary consultations by an ophthalmologist, maxillofacial surgeon, otorhinolaryngologist and a neurosurgeon. The patient ultimately was [4] brought to the operating room by an otorhinolaryngologist who removed the knife successfully under general anesthesia along the path of insertion by simple withdrawal. Intraoperatively, the soft tissue wound was explored, irrigated using normal saline and closed primarily.

Photograph 1. Showing the patient with a retained knife at the frontal region.

Photograph 2. [1–4] CT scan showing a retained knife blade at the frontal region.

Photograph 3. Showing the patient postsurgical removal of the retained knife.

Postoperatively, the patient was given intravenous ceftriaxone 1 g 12 hourly for 72 h then transitioned to oral cefuroxime 500 mg 12 hourly for 7 days, intravenous dexamethasone 8 mg 8-hourly for 72 h, transfused 2 units of whole blood (900mls), intravenous fluids (2 l of normal saline and 1 l of dextrose normal saline for 24 h), intravenous paracetamol 1 g 12 hourly for 72 h. The patient also received tetanus toxoid vaccine (0.5 ml intramuscularly). Following surgical removal of the retained knife under general anesthesia, the patient had an uneventful recovery following the scheduled postoperative follow-ups and with unremarkable sequelae apart from the wound to the left inferior rectus muscle that was managed conservatively. The patient was discharged from the hospital after 72 h.

3. Discussion

Craniofacial stab wounds secondary to retained sharp metallic objects are rarely encountered. These scenarios possess management challenges to clinicians especially when vital adjacent structures are encountered [1,2,6,7,14].

Studies have shown patients who present at emergency departments across the globe with such pattern of retained craniofacial foreign objects tend to have a high level of alcohol consumption [1,15] though contrary to what was found in our case report where no history of alcohol consumption was reported but rather assaulted by thieves while selling items at his shop.

Grobbelaar and Knottenbelt described 11 patients with retained knife blades and it was found that the knife blades could be removed along the pathway of entrance with no significant resultant sequelae. The authors suggested that impacted knife blades could safely be removed in theatre with little fear of complications that may arise upon removal [6]. Similarly in our case report, the retained knife was safely removed in theatre under GA by simple withdrawal along the path of insertion with no fearful complications.

Case series from Brazil reported knife-wounded patients to be also wounded in the thoraco- abdominal region, and some of them presented with cut wounds involving hands because of self-defense attempts. This also explains the low rate of occurrence of patients with impacted knives in the facial region, because the hands protected the face when they were assaulted [1]. This appears somehow different from what was found in our case report where the patient had no injuries involving hands during the assault due to lack of self-defense.

Some authors suggested angiography to be performed in cases where retained foreign bodies are close to major arteries. This would allow the surgeon to pre-empt the complication of acute rupture resulting from pulsation of the artery against the retained sharp object, and the subsequent development of pseudo- aneurysms. In cases of excessive arterial bleeding, angiography can help to identify a bleeding vessel that can then be managed by selective embolization or surgical ligation. If the path of the knife is clear of the skull base and major vessels, angiography is not mandatory as was found in this reported case, thus not necessitating angiography [1,6,8,10,15].

Thorough physical examination must be performed whenever encountering patients with retained craniofacial sharp metallic objects since it's important to ascertain the depth of the stab wound and integrity of neurovascular structures including facial nerve [1,2,6].

It is recommended that routine radiographic examination should be performed in all cases of deep craniofacial stab wounds and patients with a history of possible retained knife blades [1,4,8,11]. In most cases, simple removal along the entry pathway is safe and effective similar to this case report.

Studies have reported the treatment sequence for such kind of retained foreign objects to be withdrawal, wound exploration, irrigation and wound closure. This must be coupled by tetanus prophylaxis and administration of broad-spectrum antibiotics [1,2,4,7,10,11]. Similar protocol was applied in management of the patient reported in this case report and was successful.

4. Conclusion

Though rare, when an impacted knife is found involving the craniofacial region, routine radiographic examination is recommended and applies to all deep craniofacial stab wounds in case of a suspicious retained foreign body. In most cases, surgical removal along the path of insertion is often safe and effective.

Sources of funding

None.

Ethical approval

Ethical standards were reviewed and approved by the Emergency Medicine Department.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

CRediT authorship contribution statement

ZSA-Conceptualization, methodology, writing original draft.

WPL-Conceptualization and reviewing the prepared original draft of the manuscript.

OMK- Conceptualization and reviewing the prepared original draft of the manuscript.

AAK- Conceptualization, methodology and reviewing the prepared original draft of the manuscript.

Registration of research studies

N/A.

Guarantor

Dr. Zephania Saitabau Abraham takes full responsibility of the work.

Declaration of competing interest

The authors reports no conflict of interest.

Acknowledgment

Dr. Mary Jue Xu from the Department of Surgical Oncology at the University of Pennsylvania, USA is highly acknowledged for technically reviewing the manuscript especially grammar.

Provenance and peer review

Not commissioned, externally-peer reviewed.

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