Handling a Giant Cervical Aneurysmal Bone Cyst of C2 Vertebrae: Anesthetic Challenges

Case Report

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ABSTRACT

Introduction: Cervical Aneurysmal Bone Cyst (ABC), constituting approximately 2% of total spinal aneurysms, poses a distinct challenge for anesthesiologists. These benign yet locally aggressive, proliferative, and extensively osseous lesions are frequently associated with pathological fractures, significant blood loss, and positioning complexities, thereby presenting considerable difficulties in airway management. This report details the successful management of a colossal cervical aneurysmal bone cyst at our institution.

Case Report: An 18-year-old obese male (BMI 39.5kg/m^2) exhibited a progressively enlarging posterior neck swelling, limited mobility, and persistent pain. Cervical MRI revealed a 7.3x4.8cm, T2 hyperintense, multi-loculated lesion on C2 vertebra, confirmed by 3D CT scan as an ABC. Given the anticipated challenges in airway management due to a short neck with restricted movements, obesity, and the risk of aneurysmal rupture, awake fiberoptic intubation was planned but declined by the patient. We opted for endotracheal intubation using video laryngoscopy during manual inline stabilization (MILS) of cervical vertebrae. General anesthesia allowed safe bony cyst debulking in the prone position, managing intraoperative hypotension with fluid resuscitation, blood products, and vasopressors. The intraoperative course remained uneventful, and post-surgery, tracheal extubation was successfully achieved.

Discussion and Conclusion: Cervical ABCs, as rare spinal lesions, necessitate meticulous attention to airway security, concurrently addressing complications related to prone positioning, lesion rupture, acute blood loss, and cervical vertebrae fractures. A thorough pre-anesthetic assessment, coupled with a well-formulated anesthesia plan, vigilant hemodynamic monitoring, and comprehensive follow-up, contributes to optimal outcomes, particularly in navigating the intricacies of airway management.

Key Words: Acute blood loss, Aneurysmal Bone Cysts, Difficult airway management, Prone positioning, Video Laryngoscopy (VL).

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INTRODUCTION

First documented in 1934 by Jaffe and Coley, Aneurysmal Bone Cysts (ABCs) represent benign, expansive, non-neoplastic lesions characterized by multiple blood-filled cavities. Lacking endothelial cells, these lesions exhibit a distinctive composition of proliferative fibroblasts, giant cells, and trabeculae, as detailed by Park HY and Copley L[1-3]. Despite extensive research, the precise etiology of ABCs remains elusive, potentially linked to trauma, primary tumors (e.g., giant cell tumors, chondroblastoma, chondromyxoid fibrous dysplasia), other bony malignancies, or a genetic predisposition involving clonal *t*(16,17) translocation.

While ABCs predominantly manifest in the pediatric age group, with a slight female predilection, they constitute 1.4% of all primary tumors. Although commonly found in long bones such as the femur and tibia, spinal involvement occurs in 12-30% of cases, with a rarity of ~2% in the cervical spine[4-6]. DiCaprio M R noted the extensive spread of spinal ABCs, encompassing lamina, pedicles, spinous processes, vertebral bodies, and facets[6].

Clinical presentations typically involve somatic pain, neck stiffness, and swelling. Treatment modalities, ranging from surgical resection to various adjuvant therapies, have been comprehensively reviewed by Reddy K[7].

This case report addresses the formidable challenge posed to anesthesiologists when managing giant cervical ABCs. The complexity arises from the lesion's substantial size, proximity to neurovascular structures, impact on cervical stability, risk of airway compromise, and potential

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cyst rupture. Specifically, we elucidate the intricate anesthetic considerations encountered in an adolescent undergoing En bloc excision of a giant cervical ABC arising from the C2 vertebra, performed in the prone position under general anesthesia. This account underscores the critical importance of sharing insights into the management of rare and challenging cases, emphasizing the unique complexities faced during anesthesia administration for such intricate surgical interventions.

CASE PRESENTATION

We report a rare case of 18-year-old grade II obese gentleman, (weight-95kg, height-155cm, BMI of 39.5kg/m^2) diagnosed with cervical spine aneurysmal bone cyst of C2 vertebrae. Patient complained of gradually progressive swelling associated with persistent unrelenting pain in the posterior aspect of neck accompanied with difficulty in neck flexion or extension. It was not associated with any sensory or motor deficit of bilateral upper or lower limbs. There was no history of trauma, headache, loss of consciousness, vomiting, blurring of vision, seizure episodes, vertigo or difficulty in breathing. There was no past history of tuberculosis, hypertension, asthma, chronic obstructive pulmonary disease, heart disease or thyroid disorder and no history of previous surgery.

Clinical examination and airway assessment:

On his initial assessment on presentation, vitals were stable with normal general physical examination. Systemic examination of patient revealed no neurological deficits. Local examination of cervical spine revealed a hard globular swelling of 10x8cm in size at the level of C2-C5, which was fixed to the spine extending superiorly, upwards and downwards towards right side (Figure 1a). Swelling was tender and overlying skin was mobile. On airway examination, mouth opening was 3 fingers with a mallampati grade III (Figure 1b). The thyromental distance and hyomental distance were 6cm and 5cm respectively and neck circumference was 30cm. There was restriction in flexion and extension of neck along with short neck and grade 2 obesity attributing to an anticipated difficult airway and tracheal intubation.

Figure 1: Patient with large neck mass on posterior aspect of the neck (a) and airway examination with Mallampatti Grade III (b).

Pre-operative assessment and airway management:

Preoperative investigations showed Hemoglobin 12.7gm/dL, Total Leucocyte Count 9,300/mm^3, platelet count 1,66,000/mm^3, renal and liver function tests along with coagulation profile were within normal limits. MRI of cervical spine showed large 7.3x4.8cm, T2 hyperintense, multi-loculated lesion with multiple fluid levels involving posterior element of C2 vertebra, extending from C1 to C5 vertebra abutting posterior paravertebral muscles likely of aneurysmal bone cyst which was reconfirmed by 3D spiral CT scan showing an expansile lytic lesion arising from posterior aspect of C2 vertebrae. There was no significant evidence of disc protrusion leading to nerve root compression (Figures 2,3).

In view of anticipated difficult airway; awake fiberoptic intubation was planned initially but could not be executed due to patient refusal and negative consent. Endotracheal intubation following induction in manual inline stabilization (MILS) with video laryngoscopy was performed.

Figure 2: Spiral Computer Tomography Scan of Cervical Spine (3D) showing expansile lytic lesion arising from posterior aspect of C2 vertebrae.

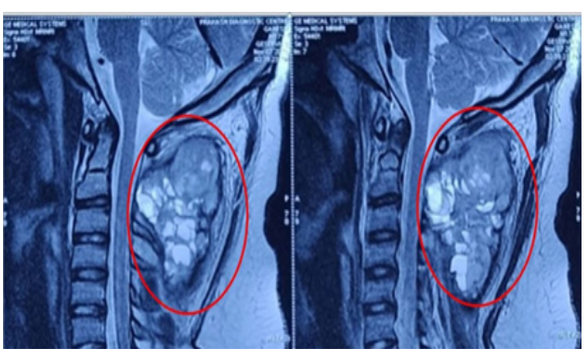

Figure 3: MRI scan of cervical vertebrae showing large T2, hyperintense, multiloculated lesion with multiple fluid/blood filled levels involving posterior element of C2 vertebra measuring 7.3x4.8cm extending from C1 to C5 vertebra abutting posterior paravertebral muscles.

Perioperative course:

Patient was taken up for surgery in the operating room with cervical collar in-situ. All Standard ASA monitors were applied along with BIS (Bispectral index system) monitor. An 18G Peripheral IV cannula and right radial arterial line was secured for invasive blood pressure monitoring. The patient was preoxygenated with 100% oxygen for 5 minutes. Premedication was done with Injection midazolam 1mg IV along with injection Fentanyl 100mcg IV followed by induction with injection propofol 120mg IV and succinylcholine 150mg IV. The trachea was intubated while maintaining manual-in-line stabilization with cervical collar in-situ with 8.0mm armored cuffed endotracheal tube using video laryngoscope which was confirmed by capnography (Figure 4a). Injection vecuronium was administered intermittently during the surgery. Patient was put on volume control mode with a tidal volume 6ml/kg body weight, Respiratory rate of 16/min, I: E of 1:2 and FiO_2 of 50%. The respiratory rate was adjusted to maintain $EtCO_2$ of around 30-35cm H_2O. Anesthesia for the patient was maintained along with oxygen and air mixture using isoflurane keeping MAC values between 0.8-1. Central venous catheter was inserted in right subclavian vein. Patient was kept in prone position with Mayfield frame and arms by the side with neck in slight flexion on Mayfield 3 pin holder (Figure 4b). Pressure points were padded.

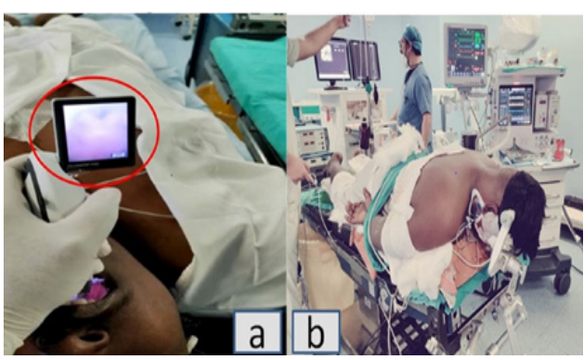

Figure 4: Video-laryngoscopic guided visualization of vocal cords (a) and prone positioning using with head supported by mayfield frame (b).

Surgical procedure:

Aneurysmal bone cyst identified using fluoroscopy, which was originating from C2 lamina with extension from C1 to C5 vertebral levels abutting posterior vertebral muscles along with bony mass had trabecular septations inside with cystic cavities filled with blood (Figure 5). Piecemeal debulking of cyst was done. Post procedure C-arm lateral view was taken to ensure adequate excision followed by CT scan (Figure 6).

Intraoperative hemodynamic instability and its management:

There were multiple episodes of hypotension during the surgery. First episode was observed during prone positioning when Invasive blood pressure (IBP) recorded was 80/55mm of Hg. Further episodes were recorded during resection of ABC. Lowest IBP recorded during surgery was 76/48mm of Hg. Hypotensive episodes were managed with IV fluid infusion initially along with intermittent vasopressor supports (noradrenaline) started with 0.05mcg/kg/min to 0.15mcg/kg/min. Total blood loss was recorded to be around 900-1000mL The blood loss was managed with one unit of PRBC (Packed Red Blood Cell) transfusion once the hemostasis achieved.

Total duration of surgery was 3 hours. Two thousand five hundred mL of crystalloids were infused intraoperatively. Urinary output at the end of surgery was around 900mL. Reversal of residual neuromuscular blockade was done with injection neostigmine and injection glycopyrrolate. Trachea was extubated after meeting adequate extubation criteria. Patient remained haemodynamically stable in postoperative period.

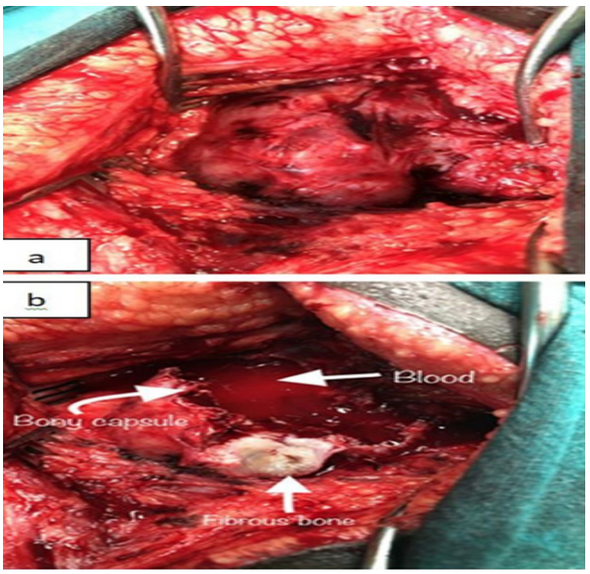

Figure 5: Trabecular septations with cystic cavities filled with blood (a) with arrow showing blood filled cavities and bony part (b).

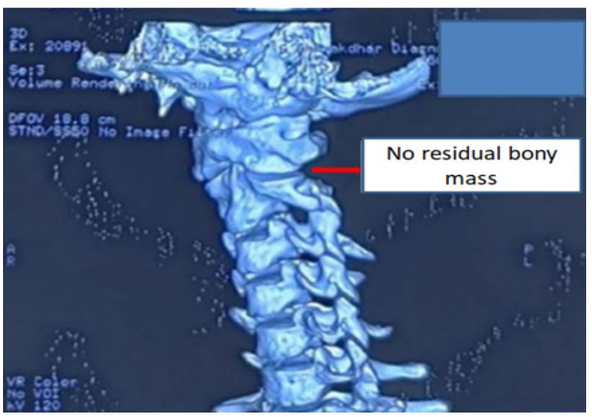

Figure 6: Post-operative CT scan showing no residual bony mass.

DISCUSSION

Aneurysmal bone cysts are non-neoplastic vascular bony lesions. According to Mehlman C T annual prevalence of such lesions is approximately 1.4 per 100,000 populations[8]. Although benign, it can aggressively expand and destroy bony tissue. ABCs most commonly occur around the knee; however it involves spine in about 12-30% of cases. The most common site affected in spine is lumbar (40-45%), and less commonly lesions are seen in thoracic and cervical spine. It is a progressively growing lesion which usually involves the posterior elements of vertebra, pedicles, body and can even extend into the spinal canal. Symptoms include swelling, tenderness, ill-defined somatic pain and stiffness. Rarely it may present with pathological vertebral fractures and neurological deficits as mentioned in earlier literature by Chan MS and Kransdorf MJ.[9,10].

Preoperative investigations and optimization:

Radiological evaluation with the help of MRI and CT scan is essential for assessment of pedicle and vertebral body integrity and thereby useful in planning the surgery. The pathognomic radiological feature is a ballooning, multilocular lytic lesion in the form of characteristic soap-bubble appearance with fluid-fluid levels. Management options include arterial embolization, radiation and surgical resection. En-bloc excision is the only treatment option that has no probability of local recurrence and its role has been well established. Intraoperative bleeding is common in ABC. Preoperative selective embolization as described by Rossi G should be considered to reduce the risk of intraoperative bleeding[11,12].

The anesthetic challenges faced in such cases can be difficulty in airway handling and management, prone positioning and its complications as well as high risk of intraoperative blood loss.

Intubation difficulty and review of literature:

We anticipated a difficult airway due to short neck of the patient, grade II obesity and restriction in flexion and extension of the neck. We therefore intubated the patient using video-laryngoscope with MILS. Excessive motion of cervical spine during intubation can precipitate rupture of aneurysm, thus further compromising airway or cord injury, as patients with ABCs are prone for cervical spine instability. MILS is preferred during intubation to reduce cervical spine mobility intubation in patients at risk of cervical spine instability as Hastings R H, noticed that secondary injuries can be prevented with cervical spine stabilization manoeuvers during laryngoscopy[13]. It has been reported that MILS reduces head extension during direct laryngoscopic intubation by 4-5° as well as upper cervical spine motion by 4° during video-laryngoscopy[14,15]. Previous studies done by Cordovani D and Suppan L have shown significant reduction in cervical spine mobility while using video-laryngoscopes for intubation than a direct laryngoscope comparing cervical spine motion during intubation between direct laryngoscopes. Also the peak and average force required to lift the epiglottis during intubation gets significantly decreases with use of video-laryngoscope than a direct laryngoscope. Therefore, video-laryngoscope provides a better laryngeal view and facilitates intubation while reducing cervical spine motion similar to previous studies[16,17].

Blood loss, hypothermia and transfusion related problems:

There is enough evidence of risk of greater perioperative blood loss during spine surgeries; osteotomies and stripping muscles off bone causing enhanced bleeding from exposed surfaces[18,19]. Intra-abdominal pressure (IAP) and relative position (Trendelenburg position) of surgical site to right atrium are important considerations in decreasing intraoperative blood loss. Hence abdomen should be freely suspended during prone positioning so as to decrease IAP and allowing drainage of blood away from the surgical site. Maintenance of normothermia is an important step for prevention of intraoperative blood loss due to hypothermia-induced coagulopathy. There is high risk of complications and mortality associated with perioperative hypothermia. Warm Intravenous fluids, warm irrigation fluids and forced air warming blanket was used for prevention of hypothermia. Verma *et al.,* in their study done on scoliosis patients on mean arterial pressure have noticed that keeping MAP <65mmHg during surgery can reduce blood loss in spine deformity cases[20]. However it can lead to decreased perfusion of spinal cord resulting in cord injury. We therefore recommend maintaining MAP >65mmHg to prevent any likelihood of ischemic injury, by use of vasopressors and intravenous fluids.

Intraoperative hypovolemia and vasodilation may result in hypotension and subsequent spinal cord ischemia. It is therefore important to maintain adequate perfusion and blood pressure during surgery. Intravenous crystalloids are the preferred resuscitative fluid in surgical patients. Colloids are used in case of massive bleeding as they don't cross the capillary membrane and remain longer in the intravascular compartment, maintaining hemodynamic stability with a lower volume of fluids. However, excessive use of 0.9% normal saline is associated with hyperchloremic acidosis and increased risk of acute kidney injury. Also, excess colloids can cause hypersensitivity reactions, coagulopathy, and renal dysfunction. Recent evidence suggests use of Plasmalyte, a balanced salt solution which has similar electrolyte composition and osmolarity to plasma which is potentially safer with no significant effect on coagulation when compared to 0.9% normal saline and colloids.

The primary indication for blood transfusion due to acute blood loss is hemodynamic instability. Excessive

blood loss can cause critical hypo-perfusion leading to a vast array of complications such as hypovolemic shock, reduced oxygen delivery, acidosis, hypothermia, dysfunctional hemostasis and coagulopathy. The main area of focus in acute blood loss is to expand intravascular blood volume and maintain oxygen delivery to tissues. Intravenous crystalloids and colloids may expand plasma volume to maintain blood pressure initially, but in case of major blood loss, blood products are required to maintain hemodynamic stability and oxygen delivery.

Due to ongoing blood loss during surgery leading to multiple episodes of hypotension, we resuscitated the patient initially with intravenous crystalloids but due to continued hemodynamic instability, packed red blood cell (PRBC) transfusion was required which eventually stabilized the patient hemodynamically.

Prone position and associated complications:

Prone position can lead to conjunctival edema, corneal abrasions, central retinal artery occlusion, ischemic optic neuropathy due to venous congestion in optic canal, cervical neuropathy and post-operative neck pain. Brachial plexus injury is observed due to stretching of nerve roots when neck is turned to a particular side for long duration, stretching of axillary bundle when humerus is abducted more than 90 and pressure on olecranon can cause ulnar nerve injury. Abdominal compression can lead to increased IAP which hinders ventilation and increases pressure in the vertebral venous plexus causing more blood loss. There can be sequestration of blood in the dependent body parts and compression of IVC which can cause hypotension due to preload reduction the heart. Direct pressure injuries can cause injury to pinna, femoral neurovascular bundle, genitalia; skin necrosis and tracheal compression. Indirect effects of increased pressure can manifest as macroglossia and oropharyngeal swelling which can lead to delayed extubation, visceral ischemia involving liver and pancreas and compartment syndromes.

In this case, video-laryngoscopic guided intubation with MILS helped us preventing life threatening complications due to minimal airway manipulation. Intraoperative hemodyanamic parameters were maintained while positioning and during acute blood loss were managed using crystalloids and blood products; keeping special emphasis on preventing position related complications in our case due to obesity.

CONCLUSION

Cervical Aneurysmal Bone Cysts are one of the rare blood filled bony lesions that are usually diagnosed in pediatric or early adult age groups. Various modalities have been described since long for the treatment of such abnormal bony cysts but En-Block excision under general anesthesia in prone position remains the treatment of choice. Such benign lesions should be carefully managed by an anesthesiologist as to minimize airway related complications, fracture of cervical vertebrae, sudden blood loss and positioning related complications. Careful pre-anesthetic check-up followed by well-conceived plan of anesthesia with hemodynamic monitoring and adequate follow-up results in better outcome.

CONFLICT OF INTERESTS

There are no conflicts of interest.

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