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Case Report

Post-Tonsillectomy Fulminant Burkholderia Sepsis at High Altitude; A Rare Complication of a Common Surgery: A Case Report

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Abstract

This case reports a 30-year-old immunocompetent male residing in high altitude region (3500 meters), who experienced severe post-tonsillectomy systemic infection of *Burkholderia cepacia* including septicaemia, multiorgan dysfunction syndrome (MODS) and disseminated intravascular coagulation (DIC). Aggressive management was necessary, emphasizing the importance of vigilant monitoring and prompt intervention in post-tonsillectomy care to address significant bleeding and bacterial infections effectively.

Keywords: Multiorgan Dysfunction Syndrome (MODS); Disseminated Intravascular Coagulation (DIC)

Introduction

Tonsillectomy is one of the most routinely performed surgeries by otorhinolaryngologists globally. Though usually performed on paediatric population, significant number of adults also undergo tonsillectomy; the most common indications in adults being chronic tonsillitis, recurrent tonsillitis, obstructive sleep apnoea and tonsillar masses requiring excision biopsy [1]. Although generally a safe surgery, surgeons should be cognizant of it's potential complications and be prepared to manage them. The most serious complication of tonsillectomy is post-operative haemorrhage. This case presents a rare severe post-tonsillectomy haemorrhage and systemic infection of *Burkholderia cepacia* resulting in septicaemia, MODS and DIC at high altitude.

Case Report

A 30-year-old male with no comorbidities presented with recurrent tonsillitis to a secondary care health center. He was residing in high altitude region (3500 meters) for the past 1 year, and was acclimatized with appropriate physiological changes. Preoperative assessment revealed normal vital parameters, grade-III tonsillar enlargement without any active infection (Figure 1) and normal systemic examination.

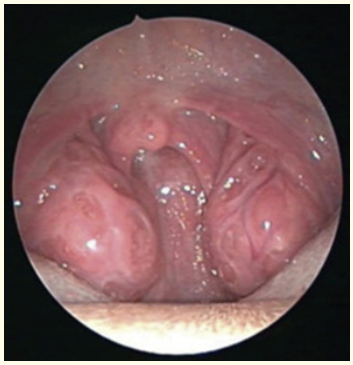


Figure 1: Clinical examination of the patient showing bilateral grade-III tonsillar enlargement.

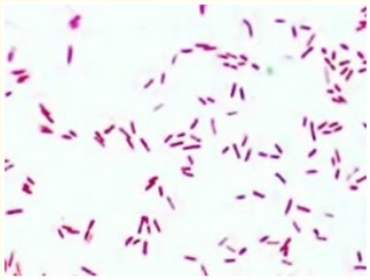
He was accepted for administration of GA under ASA-I during pre-anaesthetic checkup and underwent bilateral cold steel tonsillectomy with no intra-operative complications. Haemostasis was confirmed post-operatively before shifting him to the ward uneventfully. Within 4 hrs of surgery, he developed acute onset oozing of fresh blood from bilateral tonsillar beds. Given the large volume haemorrhage, he was returned urgently to the operating room and haemostasis was attempted with electro-cautery. However, only partial control was achieved leading to loss of approximately 800 ml of blood. Bilateral tonsillar fossae were packed and the patient remained intubated on mechanical ventilator (MV) with pyrexia and rapid deterioration of laboratory parameters (Table 1).

Investigation	Pre-op	Post -op evening	Post Operative Day (POD)-1	POD-2
Haemoglobin (g/dl)	18.1	14.9	9.3	7.6
Total leucocyte count (/uL)	4.29 x10^3	11.68 x10^3	17.3 x10^3	21.6 x10^3
Platelets (/uL)	2,57,000	71,000	38,000	30,000
Serum urea (mg/dL)	22.8	48	53.7	84
Serum creatinine (mg/dL)	0.94	1.76	2.4	3.1
Serum bilirubin (mg/dL)	0.8	4.78	6.33	5.5
SGOT (IU/L)	65	298	424	434
SGPT (IU/L)	30	161	203	294
C- reactive protein (mg/dL)	-	Negative	Positive	12.5
Fibrin degeneration products (mg/L)	-	Positive	Positive	176
Serum ferritin (ng/mL)	-	-	1437	2249
Serum procalcitonin (ug/L)	-	-		69.8
Fibrinogen (mg/dL)	-	-	290	194
LDH (IU/L)	-	-	3940	1004
D-dimer (ng/mL)	-	-	Positive	>10,000
Chest X-ray	Normal	Normal	Normal	Bilateral lower zone consolidation

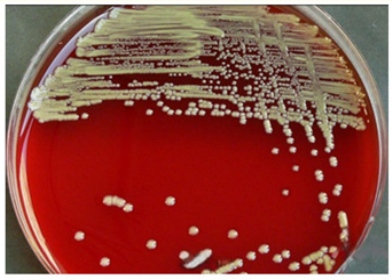
Table 1: Laboratory Parameters.

He was diagnosed as septicaemia with DIC and MODS and transferred to a tertiary care center where he was detected to have acute pyelonephritis, pneumonia and deranged liver functions. Microbial assessment revealed colonies of gram-negative rods on gram staining which were motile and tested positive for catalase and oxidase production and also for nitrate reduction (Figure-2a). They also oxidized sugars (glucose, lactose, mannitol, and maltose). They decarboxylated lysine but did not hydrolyze arginine. Blood samples were sent for culture on two separate occasions where both times, the culture showed pure growth of non-lactose fermenting, mucoid, smooth colonies with diameter of approximately 2 mm (Figure-2b). The antibiotic susceptibility test showed that the organism was multi-drug resistant and only intermediately susceptible to Amikacin (Figure-2c). The organism was subjected to identification by automated VITEK2 Compact system and was identified as *Burkholderia cepacia*.

a



b



c

Identification Information		**Analysis Time:** 3.35 Hours		**Status:** Final	
Selected organism		98% Probability **Burkholderia cepacia** Bionumber: [redacted]		**smooth and raised colonies**	
ID Analysis Messages					
Susceptibility Information		Analysis Time: 17.27 hours		Status: Final	
Antimicrobial	**MIC**	**Interpretation**	**Antimicrobial**	**MIC**	**Interpretation**
Piperacillin/ Tazobactam	>= 128	R	Amikacin	32	I
Ceftazidime	>= 64	R	Gentamicin	> 16	R
Cefoperazone/Sulbactam	>=64	R	Ciprofoxacin	> 4	R
Cefepime.	>=32	R	Levofloxacin	> 8	R
Aztreonam	>= 64	R	Minocycline	>= 32	R
Imipenem	>= 16	R	Trimethoprim/ Sulfamethoxazole	>= 320	R
Meropenem	>= 16	R			

Figure 2a: Burkholderia cepacia seen as gram-negative rods.

Figure 2b: Burkholderia cepacia on blood agar showing smooth and raised colonies.

Figure 2c: Blood for Culture and Sensitivity Report.

Immunocompetency workup including HIV, hepatitis-A, hepatitis-B and hepatitis-C status failed to reveal any immunocompromised state. Histopathological examination of tonsils showed reactive lymphoid hyperplasia, ruling out tonsillar malignancy. He remained on MV, aggressive management was done with administration of intravenous Amikacin, Piperacillin-Tazobactam and Teicoplanin for culture driven as well as broad spectrum coverage for any secondary infections. Blood component replacement with fresh frozen plasma and haemocoagulase local pack application in tonsillar fossae was done. Injectable Vitamin K and Tranexamic Acid were also administered. On POD-4, he showed recovery in form of cessation of tonsillar bed ooze and improvement of laboratory parameters. He was extubated on POD-6, recovered completely by POD-15 and discharged. Follow up after 6 months revealed normalized lab investigations and no immunocompromised state.

Discussion

Tonsillar bed haemorrhage following tonsillectomy is a significant post-operative complication in 1-4% cases despite adequate intra-operative haemostasis with a potential to cause serious morbidity and death. The mortality rates vary from 1/10,000 to 1/ 35,000, mainly as a result of haemorrhage [2]. Windfuhr., *et al*. reported mortality from uncontrolled haemorrhage in 2/ 10,000 tonsillectomies, most cases in the first 24 hours post-operatively [3]. Crysdale and Russel observed that 76% of primary haemorrhage occurred during first 6 hours following tonsillectomy; recommending monitoring in the early 6–8 hours [4].

Fulminant sepsis is a rare complication of tonsillectomy. Although various mechanisms have been postulated, it's pathophysiology remains controversial. Contrary to most surgeries where surgical wounds are sutured primarily, tonsillar bed wounds heal by secondary intention post tonsillectomy. The colonisation of pathogens in this open wound during and after tonsillectomy acts as foci of infection. Studies attribute bacteraemia either to the retrograde flow of bacteria through exposed vessels or to operative site manipulation like traction and handling of tonsil by forceps before or during dissection, gauze tamponade application or suture ligation following intra-operative haemorrhage or overpressure to tonsillar fossa while suctioning as the route of entry of microorganisms into systemic circulation [5]. This bacteraemia is usually transient, however it may lead to dramatic results in immunocompromised patients. The commonly isolated organisms in blood cultures from patients with post-tonsillectomy septicaemia are Invasive group A streptococcus, Staphylococcus aureus, Haemophilus influenzae and anaerobic bacteria, usually similar to the isolated bacteria from the tonsillar surface. Severe group A beta haemolytic streptococcal infection and even death after uncomplicated tonsillectomy have been reported in many studies [6,7]. Septicaemia by *Burkholderia cepacia* following tonsillectomy is rarely reported. However, Burkholderia cenocepacia was demonstrated in tonsil specimens of two immunocompetent patients following uncomplicated tonsillectomy for recurrent tonsillopharyngitis by Zautner., *et al*. [8].

Burkholderia cepacia is a ubiquitous aerobic gram-negative bacillus. Isolated from soil, water and plants, a group of nine genetically related non-fermentative bacteria are collectively termed as *Burkholderia cepacia* complex: *B. cepacia, B. multivorans, B. cenocepacia, B. stable, B. vietnamiensis, B. dolosa, B. ambifaria, B. anthina* and *B. pyrrocinia*. *Burkholderia cepacia* rarely cause disease in normal host. It mostly causes life-threatening respiratory infections in cystic fibrosis patients, however may seldom also lead to post-operative nosocomial infections like bacteremia, meningitis, endocarditis, peritonitis, or UTI in immunosuppressed patients and/or in those with significant comorbidities. To mention a few, a case of B. cepacia septicemia was reported in a patient of rheumatic heart disease with pleural effusion who had undergone mitral valve replacement [9]. Khoa., *et al*. illustrated hospital-acquired pneumonia due to *Burkholderia cepacia* in a thalassemia pregnancy with postoperative eclampsia [10].

Only a handful cases of post-operative manifestations of *Burkholderia cepacia* in immunocompetent patients have been reported in literature. A rare case of *Burkholderia cepacia* infection was described in an immunocompetent patient following pancreaticoduodenectomy [11]. Iatrogenic postoperative spondylodiscitis attributed to *Burkholderia cepacia* infection in an immunocompetent patient was reported by Rakesh., *et al*. [12]. Weinstein., *et al*. elucidated a case of cervical osteomyelitis caused by *Burkholderia cepacia* after an elective rhinoplasty [13]. However, none has been described following tonsillectomy to the best of our knowledge.

In hospital settings, common sources of *Burkholderia cepacia* are vascular and indwelling catheters. *Burkholderia cepacia* has been implicated in outbreaks from contaminated surgical compounds [14,15] as well as pharmaceutical products such as nebulization solutions, hand sanitizers and mouthwash [16,17]. Postoperative endophthalmitis due to *Burkholderia cepacia* complex from contaminated anaesthetic eye drops was highlighted by Lalitha., *et al*. [18] In the present case, operating room swabs, products used for preoperative skin antisepsis, iv fluids used for irrigation/gauze

tamponade, drugs used for tonsillar bed tamponade intra-operatively and intravenous catheters were tested for any contamination but none was found.

It is pertinent to consider the effect of high altitude as a confounding factor in our case. High altitude environments can exacerbate or contribute to the symptoms of recurrent tonsillitis, primarily due to environmental stressors like dry, cold air and low humidity, which impair the natural defenses of the upper respiratory tract mucosa; especially in the non-native residents as in our case. The low relative humidity and cold temperatures at high altitudes lead to dryness and crusting of the nasal and pharyngeal mucous membranes making the throat more susceptible to infections [19]. In addition, nasal obstruction, a common issue at high altitude due to the dry air, often leads to mouth breathing. This bypasses the nose's natural function of warming and moistening the inhaled air, further drying out the throat and increasing irritation. The dry, cold conditions and hypoxia tend to impair the normal mucociliary clearance mechanisms, increasing the risk and severity of respiratory tract infections, including tonsillitis [20]. Exposure to high-altitude hypoxia can induce local inflammation and affect the balance of oral microbiota, potentially making the tonsils and surrounding tissues more vulnerable to pathogens. While high altitude does not introduce new specific pathogens, the harsh environmental conditions and the body's physiological responses can weaken local defenses and increase susceptibility to the common infections that cause recurrent tonsillitis [21].

Hypoxia-induced compensatory hyperplasia of red blood cells and the increase in blood viscosity in response to high altitude accelerate the consumption of coagulation factors, increase risk of thrombogenesis, and ultimately prolong prothrombin time and activated partial thromboplastin time and low fibrinogen levels. High altitude also affects platelet aggregation or adhesion. Fibrinolysis is a normal physiological process that controls where fibrin clots form and how large they grow and that removes clots during healing. Low fibrinogen levels destabilize clots by making them more susceptible to fibrinolysis, which can lead to uncontrolled bleeding. This phenomenon may be a physiological compensatory response to the hypercoagulable state induced by high altitude which could have been the case in our patient as well [22]. In a study conducted by Zhao., *et al*. high altitude was associated with greater total blood loss and hidden blood loss after total knee arthroplasty; longer activated partial thromboplastin time, prothrombin time, and thrombin time; significantly higher FDP levels substantiating the increased risk of operative and post-operative haemorrhage in surgeries performed at higher altitudes [23].

In the present case, whether septicaemia was a post-tonsillectomy complication triggered due to the extreme environmental conditions, an iatrogenic infection or post-operative contamination needs deliberation. The exact cause of septicaemia in this case could not be substantiated with certainty despite best efforts, as it was an isolated case. Early onset of septicaemia within few hours of surgery points towards intra-operative or early post-operative dissemination which could be from the tonsillar surface.

Conclusion

Post-Tonsillectomy bleeding is a rare, yet difficult condition to manage. In this case, post-tonsillectomy haemorrhage at a high altitude setting as a result of fulminant septicemia with DIC and MODS caused by a rare pathogen posed several challenges highlighting the need for vigilant post-operative monitoring and prompt intervention for successful management of rare complication of a common surgery.

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