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CASE REPORT

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Rare case of necrotizing tonsillitis causing severe airway infection in an infant: a case report

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Abstract

Background Necrotizing tonsillitis is rare and may lead to life-threatening upper airway obstruction in children, requiring emergency airway management.

Case presentation An 8-month-old boy presented with unresolved fever and was diagnosed with acute tonsillitis. Despite prior treatment with amoxicillin and paracetamol, the fever persisted, accompanied by leukopenia. Intravenous C-penicillin was initiated, but respiratory distress ensued, necessitating non-invasive ventilatory support and subsequent intubation due to increased stridor. Intubation was challenging due to copious secretions and a floppy epiglottis, but successful intubation was achieved on the second attempt using a C-MAC® video laryngoscope with a Miller blade size 0. Computed tomography (CT) revealed a large collection with mucosal involvement in the peritonsillar and tonsillar regions, extending to adjacent structures. Direct laryngoscopy, tissue sampling, and multiple surgical debridements were performed as the patient's condition deteriorated. Perianal excoriation and diarrhea raised suspicion of primary immunodeficiency syndrome. *Pseudomonas aeruginosa* and *Stenotrophomonas maltophilia* were isolated from tissue cultures and effectively treated with targeted antibiotics. Serological testing showed positive IgG for herpes simplex virus 1 (HSV-1), while immune deficiency testing indicated a normal immune status, pending genetic testing results. After 21 days of ventilation, the patient was extubated, received non-invasive ventilatory support, and was discharged with oral antibiotics.

Conclusion This case highlights the critical nature of necrotizing tonsillitis, especially in infants with suspected primary immunosuppressive disorders. Early recognition, prompt airway management, and surgical intervention are crucial for optimal outcomes.

Keywords Airway, Obstruction, Necrotizing tonsillitis, Infant, Immunosuppression, Case report

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Background

Necrotizing tonsillitis is a rare and severe form of tonsillitis characterized by necrosis and inflammation of the tonsil tissue. It is most commonly seen in adults with underlying immunosuppressive conditions, such as HIV, leukaemia, or diabetes mellitus. However, it can also occur in previously healthy individuals, particularly infants and young children. Viral infections that cause tonsillitis and pharyngitis are common and usually self-limiting. These viruses include rhinovirus, respiratory syncytial virus, Epstein–Barr, parainfluenza and influenza, coxsackievirus, adenovirus, as well as the herpes simplex virus (HSV) [1]. Upper respiratory tract infection may result in acute tonsillitis associated with tonsillar hypertrophy, which may progress to serious complications, including necrotizing tonsillitis with or without airway compromise, especially in an immunocompromised host.

Tonsillopharyngitis is an inflammation of the tonsils and structures of the pharynx, including Waldeyer's ring, compromising the lingual and palatine tonsils as well as the adenoids. Infection may extend to the epiglottitis or result in abscess formation. However, it rarely progresses to life-threatening airway compromise. Tonsillar enlargement occupying more than 75% of the oropharyngeal inlet can cause severe airway compromise, even though the patient may appear misleadingly calm [2]. The lingual tonsils are notorious for causing unanticipated airway obstruction during anaesthesia as infection may be asymptomatic and undetected in routine airway assessment. In this report, we describe an extremely rare and severe case of upper airway obstruction in an infant due to necrotizing tonsillitis.

Case presentation

Written consent for the publication of this case report was obtained from the parents of the infant. An 8-month-old boy presented with unresolved fever and poor oral intake despite a 5-day treatment with amoxicillin and paracetamol syrup for tonsillitis. He was born full-term, had achieved normal developmental milestones, and had up-to-date immunization. On presentation, the patient was lethargic with mild dehydration. His heart rate was 170 bpm and a blood pressure of 107/55 mmHg. Tachypnea was identified at 33 breaths/min, his temperature was 38.2 °C, and his oxygen saturation was 100% on room air. The patient's throat was inflamed; however, there was no ulcer or exudate with hypertrophic tonsils. Blood tests showed normochromic normocytic anaemia (haemoglobin 10.8 g/dL, mean corpuscular volume 76.0 fL, mean corpuscular haemoglobin 24.6 pg), leukopenia (total white cell counts 4.24×10^9/L), and raised inflammatory marker levels (C-reactive protein 101.04 mg/dL) (Table 1). A provisional diagnosis of acute tonsillopharyngitis was made. The patient was immediately admitted and commenced on intravenous (IV) benzylpenicillin 285,000 IU four times a day (50,1000 IU/kg/dose). Screening for COVID-19 infection was negative. Overnight, the high-grade fever persisted, he developed watery diarrhoea, and his clinical status deteriorated rapidly. A loud stridor and drooling were present, and the infant preferred to be in the left lateral position with his neck extended. Reassessment showed bilateral tonsils grade 3, erythema with exudate, and perianal excoriation, which raised the suspicion of acute epiglottitis and retropharyngeal abscess with underlying primary immunodeficiency syndrome. Peripheral blood was sent for lymphocyte subset flow cytometry, Nitroblue tetrazolium (NBT) and DNA testing in order to investigate the infant's immune status prior to an administration of IV immunoglobulin, while the antibiotic therapy was escalated to IV ceftriaxone, cloxacillin and gentamicin. He became more tachypneic and dyspneic and required non-invasive ventilation (NIV) support. A referral to the oto-rhinolaryngology and anesthesiology teams was initiated and, an urgent neck computed tomography (CT) was

Table 1 Range of infective markers during the hospital stay

Investigation	On admission	HDU admission	Before CT scan	Before 1st ENT Procedure	Before 2nd CT scan	Before 2nd OT	Before extubation	Discharged from PICU	Reference value
Haemoglobin	10.1	8.2	8	11.3	9.9	8.5	11.2	11.9	(13.0–17.0) g/dL
Total white cell count	4.24	4.62	4.65	17.65	24.29	12.84	9.39	7.13	(4–10) x10^9/L
Neutrophil count	0.53	1.99	2.15	11.22	16.08	7.65	6.46	2.41	(1.19–7.21) x 10^9/L
Leucocyte count	1.97	1.54	1.65	3.86	4.42	2.87	1.92	3.88	(1.56–7.83) x10^9/L
Monocyte count	1.73	1.02	0.82	1.93	3.63	2.19	0.91	0.52	(0.25–1.15) x10^9/L
Eosinophil count	0	0.01	0.01	0.29	0.13	0.1	0.04	0.26	(0.02–0.82) x10^9/L
Basophil count	0.01	0.06	0.02	0.16	0.03	0.03	0.06	0.06	(0.01–0.06) x10^9/L
Platelet	222	84	81	80	269	363	540	362	(150–410) x10^9/L
C-Reactive Protein	101.04	-	238.86	180.26	-	-	84.69	-	< 0.5 mg/dl
Procalcitonin	-	24.88	-	0.92	-	0.35	9.68	0.22	0.02–0.30 ng/ml
ESR	-	-	55	-	-	-	-	-	(1–15) mm/hr

planned followed by a direct laryngoscopy and examination under anaesthesia in the operating theatre after the airway was secured.

The infant was transported to the operating room (OR) on NIV with nasal continuous positive airway pressure (positive end-expiratory pressure [PEEP] 4 cmH_2O, fraction of inspired oxygen [FiO_2] 0.3). A difficult airway drill was initiated by the otorhinolaryngology team, anticipating the need for a surgical airway. He was preoxygenated with FiO_2 1.0, maintaining spontaneous breathing with a PEEP of 5 cmH_2O and sevoflurane 8%. IV fentanyl 2 µg/kg and succinylcholine 1.5 mg/kg were also administered. Intubation was assisted using a C-MAC® video laryngoscope (Karl Storz SE, Tuttlingen, BW, DEU) with Miller blade size 0. After initial difficulty due to copious amounts of thick secretion and an edematous, floppy epiglottis, a 3.5 mm endotracheal tube was successfully placed on the second attempt. There was a brief desaturation that immediately improved, while the hemodynamic parameters remained stable. An immediate CT scan of the neck revealed a large, bilateral, multiloculated collection with mild rim enhancement that measured 2.4 × 3.9 × 3.8 cm (AP x W x CC) and was centred at the mucosal pharyngeal space of the peritonsillar and tonsillar regions. This suggested the presence of a bilateral peritonsillar abscess with extension into the vallecular region and extensive reactive oedema/phlegmon surrounding the retropharyngeal, prevertebral, and posterolateral neck regions. Additionally, X-ray evidence showed probable aspiration pneumonia (Fig. 1).

An examination under anaesthesia with direct laryngoscopy and nasoendoscopy was performed. We observed friable slough in the oropharynx and supraglottic structures, a mucosal breach of the posterior pharyngeal wall, and an erythematous and edematous

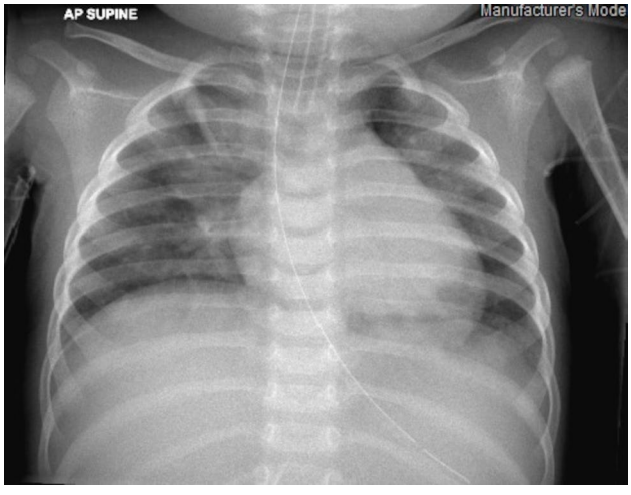

Fig. 1 Chest X-ray showing bilateral lung consolidation likely due to aspiration pneumonia

epiglottis and false cords (Fig. 2A and B). Slough was also present in the left middle turbinate, and the right nasal septum and adenoid were hypertrophic. A tissue biopsy and bilateral aspirations of the tonsillar and peritonsillar regions were performed, with only a minimal amount of material collected. Toileting and debridement procedures were also performed. Following tissue culture that grew *Pseudomonas aeruginosa*, the antibiotic treatment was changed from IV ceftriaxone, cloxacillin, gentamicin, and metronidazole to IV cefepime and amikacin (Tables 2 and 3). However, a repeat CT scan of the neck at day-10 of illness showed worsening of the multiloculated collection in the upper airway (Fig. 3). Its epicentre at the mucosal pharyngeal space of the bilateral peritonsillar and tonsillar regions had increased in size, extending to approximately 2.9 × 4.7 × 3.8 cm (AP x W x CC). The fullness of the naso-oropharynx with edematous pharyngeal mucosa caused significant narrowing of the upper airway tract. Multiple, enlarged, lobulated cervical lymph nodes with a new area of central hypodensity suggested nodes with central necrosis. At this point, histopathology of the tissue obtained at the first debridement became available and revealed an area of necrosis (Fig. 4). The diagnosis was revised to bilateral necrotizing tonsillitis. On day-11 of the illness, the patient underwent further debridement and toileting under anaesthesia (Fig. 2C). The airway remained inflamed and edematous, warranting prolonged ventilation support. The culture originating from tissue taken at the second debridement grew *Stenotrophomonas maltophilia* with sensitivity to trimethoprim/sulfamethoxazole. Viral serology screening was also performed and showed IgG-positive for HSV-1 after 10 days (Table 4). He was successfully extubated to NIV with bilevel positive airway pressure (PEEP 10/6 cmH_2O, FiO2 0.4) after 21 days of ventilation support. The patient was discharged with trimethoprim-sulfamethoxazole syrup 5 mg/kg to be administered every other day as an oral prophylaxis, considering primary immunodeficiency syndrome, which was later discontinued as the lymphocyte flow cytometry and NBT analysis showed normal immune status. The patient was followed up at the outpatient daycare, with his developmental milestones progressed. The child was alert, cheerful, not tachypneic, had good hydration status, a body weight of 6 kg (below −2 SD but increasing in trend) with primary immunodeficiency workup being normal.

Discussion and conclusion

Necrotizing tonsillitis which developed into an acute upper airway obstruction, especially in an infant is extremely rare. Nevertheless, acute tonsillitis which rapidly developed into a severe upper airway obstruction requiring emergency cricothyroidotomy has been reported in adults. This has shown how a complicated

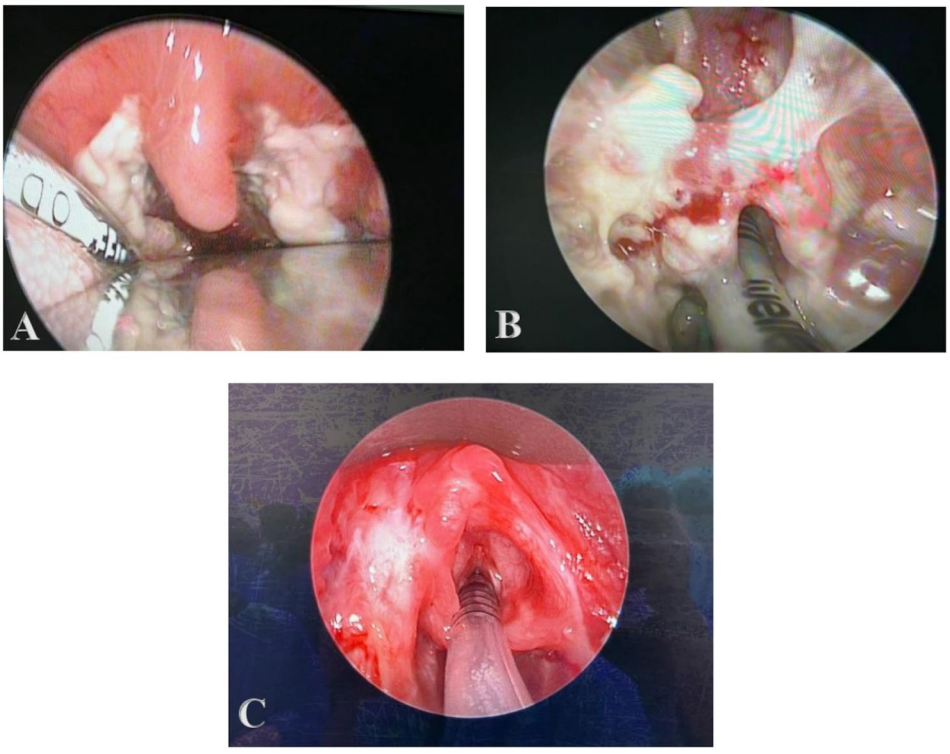

Fig. 2 Direct laryngoscopy showing (**A**) friable slough in the oropharynx and supraglottic structures; (**B**) mucosal breach of the posterior pharyngeal wall, erythematous and edematous epiglottis and false cords; and (**C**) markedly improved mucosal surfaces of the posterior pharyngeal wall, epiglottis, and false cords following multiple wound toileting and debridements

Table 2 Tissue microbiology

Sample	Culture result	Sensitivity
Tracheal aspirate (x3)	Negative for acid-fast bacilli	-
Urine	No growth	-
Peripheral blood	No growth	-
Left valleculae	*Pseudomonas aeruginosa*	Cefepime, amikacin, ceftazidime, gentamicin
Laryngeal tissue (intraoperative sample)	*Pseudomonas aeruginosa* Negative for acid-fast bacilli Negative for *Mycobacterium tuberculosis* Negative for *Corynebacterium diphtheriae*	Ceftazidime, gentamicin
Left tonsil (intraoperative sample)	*Stenotrophomonas maltophilia*	Trimethoprim-sulfamethoxazole
Right tonsil (intraoperative sample)	*Stenotrophomonas maltophilia*	Trimethoprim-sulfamethoxazole

Table 3 Summary of antibiotic prescription

Day of Hospital Admission	Tissue Microbiology and Sensitivity	Antibiotic Prescribed	Comment
Before hospital admission	-	Oral amoxicillin	-
Day 1	-	IV benzylpenicillin	Empirical antimicrobial agent for tonsillitis
Day 2	-	IV ceftriaxone, IV cloxacillin, IV gentamicin	Escalated antimicrobial therapy due to deterioration in overall clinical condition, to cover for gram negative sepsis
Day 7	*Pseudomonas aeruginosa* sensitive to cefepime, amikacin, ceftazidime, gentamicin	IV cefepime, IV amikacin	Adjusted antimicrobial therapy based on tissue microbiology and antimicrobial susceptibility testing, covering for hospital acquired infection
Day 10	*Stenotrophomonas maltophilia* sensitive to trimethoprim/sulfamethoxazole	IV trimethoprim-sulfamethoxazole	Adjusted antimicrobial therapy based on tissue microbiology and antimicrobial susceptibility testing, covering for hospital acquired infection

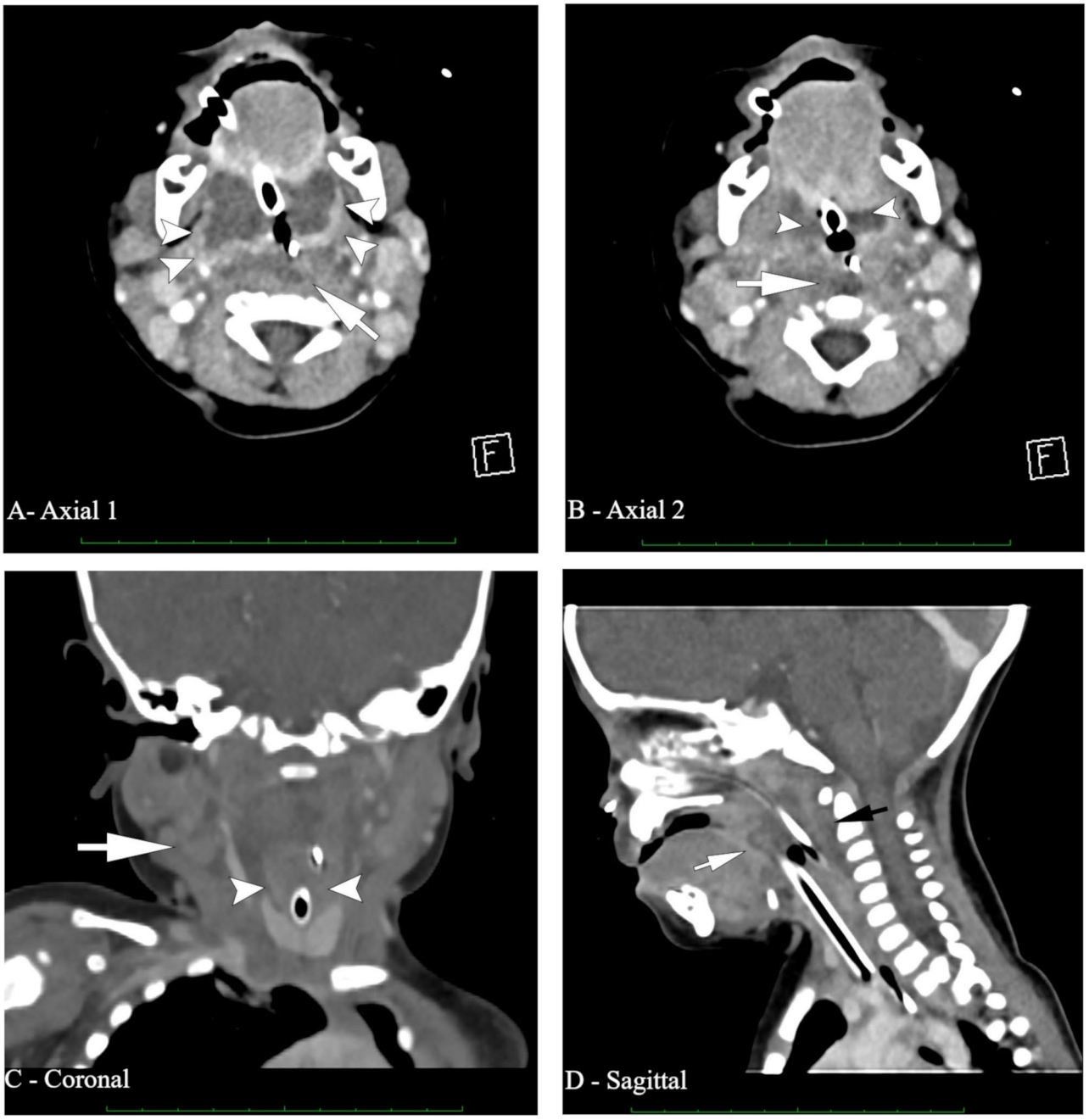

Fig. 3 (**A**) (labelled as Axial 1): Axial view at the level of tonsillar adenoid: rim-enhancing collection obliterating the lumen at the tonsillar region (arrowheads) and extension posteriorly at the level of oropharynx (arrow). (**B**) (labelled as Axial 2): Axial view at the level of base of tongue: extension of rim-enhancing collection inferiorly at the base of tongue and into the oropharynx (arrowheads) and retropharyngeal collection (arrow). (**C**) (labelled as coronal): Coronal view showing collection around the endotracheal tubing (arrow heads) with an enlarged right 1b cervical node (arrow). (**D**) (labelled as sagittal): Sagittal view showing collection in the oropharyngeal airway (white arrow) and retropharyngeal collection (black arrow)

infection may progress into acute supraglottis with narrowing of the upper airway and subsequent respiratory distress due to airway compromise [3]. In this case, the patient's obstructed airway was managed early. A trial of NIV was utilized as a bridging therapy in which definite airway support was provided in the OR by the pediatric anaesthesia team. To date, there are various approaches to managing acute airway obstruction [2, 4]. Most importantly, clinicians must be able to anticipate a potentially difficult or compromised airway. This requires an understanding of the advantages and limitations of various types of airway equipment. Although uncommon, studies have suggested the potential benefits of early debridement when treating patients with similar pathology where

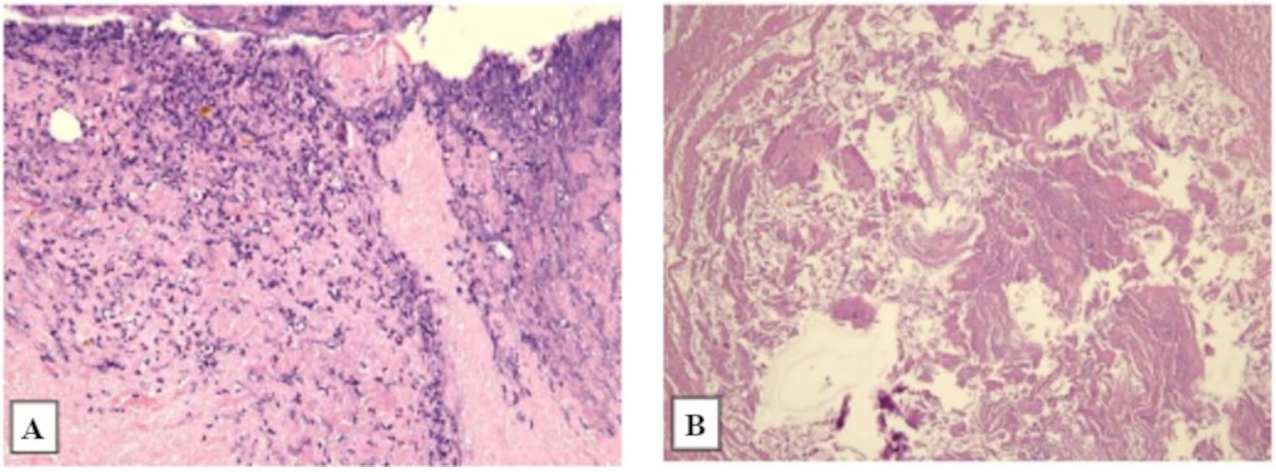

Fig. 4 Microscopic view of (**A**) the left tonsil segment, showing necrotic tissue focally covered by fibrinosuppurative exudate with neutrophils and apoptotic debris; and (**B**) the right tonsil, showing anucleate keratin debris within the tissue. No viable tissue is present and no granuloma or fungal body is observed

Table 4 Serum antigen and antibody

Serological Test	Results
Serum Aspergillus Antigen (ELISA)	Negative
Serum Candida Antigen (ELISA)	Negative
Herpes simplex virus 1&2 IgM	Negative
Herpes simplex virus 1&2 IgG	Positive

surgical intervention was considered to avoid local tissue loss, airway intervention, and long-term complications [4, 5]. Securing and maintaining the airway during every debridement and toileting procedure is challenging, as it is a shared airway surgery. Hence, thorough preparation, including having a difficult airway trolley, video laryngoscope, laryngeal mask airway, and endotracheal tubes of varying sizes at hand, was vital. Additionally, experienced pediatric anaesthetists and ear, nose, and throat specialists were present throughout the procedures.

Necrotizing tonsillitis caused by HSV-1 infection in immunodeficient hosts has been reported in adults in the past. In this infant, no history of immunosuppression was revealed; however, he was anaemic and leukopenic on presentation. HSV-1 infection of the upper airway often is asymptomatic and common in children and therefore should be considered in the differential diagnosis of immunosuppressed patients [6]. In this case, the initial blood samples for HSV-IgG were positive suggesting previous infection, or less likely maternal origin. Highly specific or sensitive testing for HSV such as culture, polymerase chain reaction or direct immunofluorescence assay was not performed due to the lack of laboratory facilities in our centre. Although histopathological findings are useful in confirming an infection, the necrotic tissue section in our case did not show a herpetic pattern. Due to this, antiviral therapy was not prescribed for this infant.

The diagnosis of necrotizing tonsillitis is often difficult due to the rarity of the condition and the nonspecific nature of the symptoms especially in an infant with a suspected primary immunodeficiency. The infant in this case had been treated with antibiotics prior to admission; however, his condition worsened and led to respiratory distress. During the first laryngoscopic examination, both tonsils and the oropharynx were inflamed and edematous, with no observed exudate or necrosis. In cases of no response to the first-line regimen after 72 h, the current recommendations, which we followed, proposes changing the antibiotic and continuing to investigate the cause using microbiological cultures, serology, or other laboratory molecular techniques [7]. The antibiotic regimen was revised to treat the *P. aeruginosa* and *S. maltophilia* infections identified in tissue cultures. Pseudomonas infection is known to potentially lead to necrosis. Surgical intervention with multiple toileting and debridement of the tonsillar and peritonsillar regions was required due to the severity of airway obstruction and the patient responded well to surgical intervention and targeted antibiotic therapy.

In summary, necrotizing tonsillitis should be considered in the differential diagnosis of infants with severe airway obstruction and tonsillitis. Early recognition and prompt treatment, including airway management and surgical intervention, are crucial for a successful outcome.

Abbreviations

CT	Computed tomography
HSV	Herpes simplex virus
IV	Intravenous
NBT	Nitroblue tetrazolium
NIV	Non-invasive ventilation
OR	Operating room
PEEP	Positive end-expiratory pressure

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Author contributions
FHA and LYX conceptualized and drafted and wrote the manuscript. FHA, LYX and MIA acquired, analyzed and interpreted the data and images. RT, MNM and IA edited the manuscript content and provided writing assistance. FHA, LYX and AI critically revised and proofread the manuscript. All authors approved the final version of the manuscript.

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Data availability
The data from the current study are available from the corresponding author on reasonable request.

Declarations

Ethics approval and consent to participate
Not applicable.

Consent for publication
Written informed consent was obtained from the parents of the infant involved in the study.

Competing interests
The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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