Necrotising fasciitis of the neck: Unusual presentation with aggressive management – case report with review of literature

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ABSTRACT
Tonsillitis and peri-tonsillar abscess are common conditions which account for a large proportion of hospital admissions. They may present in a similar way to supraglottitis and retropharyngeal collection. With adequate antibiotic coverage, admissions for tonsillitis have decreased compared to cases admitting with complications. This report shows an unusual presentation of necrotising fasciitis in the neck of a 71-year-old male who presented initially with tonsillitis and later developed into supraglottitis with retro- and parapharyngeal abscess complicated by necrotising fasciitis extending into pleural cavity. The patient required a prolonged stay in the Intensive Care Unit, with airway support including tracheostomy, and underwent repeated surgical drainage followed by sequential debridement (10 times). Radiological investigation and nasopharyngolaryngoscopy plays an important role in contributing to early diagnosis. Aggressive debridements and necrotic tissue removal with antibiotic cover are mainstay of treatment.

INTRODUCTION
Necrotising fasciitis (NF) is a rare and life-threatening infection which spreads along the fascial planes and subsequently causes necrosis of subcutaneous fat, skin and sometimes muscle.[1] Infection primarily involves the superficial fascia and progresses into surrounding tissues; progress is often fulminant and may involve all soft-tissue components, it can occur after inadequate care of abscesses. Predisposing factors include diabetes, obesity, peripheral vascular disease, chronic renal failure, poor nutritional status, smoking or old age.[1] Head and neck is usually less common site for NF compared to limbs. It is usually secondary to dental, peritonsillar infection which can progress to para- or retropharyngeal abscess and to mediastinum.[2] Retropharyngeal abscess caused by supraglottitis is rare. Presenting features may vary from generalised erythema, oedema of neck or may progress rapidly to shock-like stage.[3]

CASE REPORT
A 71-year-old male was referred to emergency ENT service with a one week history of sore throat and increasing difficulty with swallowing. Other than feeling generally unwell, he had no other symptoms, such as weight loss or fever, neck swelling, reduced mouth opening (trismus) or restriction of neck movements. Breathing was not restricted. He had a background history of previous colorectal cancer, atrial fibrillation and mild mitral regurgitation; he was taking Bisoprolol, Aspirin and Lansoprazole. Otherwise, he usually kept well with no reduction in exercise tolerance.

On initial examination on the night of admission, he was found to have bilaterally enlarged tonsils with surrounding palatal oedema. A diagnosis of tonsillitis was made and he was admitted following the protocol of analgesia, intravenous fluid resuscitation and i.v. antibiotics (i.v. amoxycillin). Vital signs were monitored throughout.

Despite good antibiotic cover, there was no improvement in his symptoms over the next two to three days. A flexible nasopharyngolaryngoscopy on second day of admission revealed a swollen epiglottis with surrounding symmetrical oedema with normal finding on the posterior pharyngeal wall; the diagnosis of supraglottitis was made. The treatment plan was...
amended according to the microbiologist (gentamycin + metronidazole added to amoxicillin). Regardless of these measures, the swallowing further deteriorated and the patient developed swelling around the submental and submandibular areas on both sides. Ultrasound examination showed midline swelling, suggestive of oedema and inflammation of the subcutaneous tissues with no obvious fluid/abscess collection identified.

Nasogastric feeding was commenced to improve nutritional status of the patient. Five days following this, respiratory distress was developed with continuous fall in saturations despite oxygen. He was then transferred to Intensive Care Unit for CPAP and was subsequently intubated, ventilated and managed for septic shock. See Table 1 for laboratory results throughout the disease.

Owing to worsening of his condition during nine days of admission and antibiotic cover, he underwent CT scan neck and thorax (Figures 1 and 2) which showed a large retropharyngeal abscess extending from skull base to C7/T1, right-sided pneumonia and large right-sided pleural effusion.

He was taken to theatre for trans-oral surgical drainage of the retropharyngeal abscess which revealed presence of smelly fluid with small amount of pus. The chest drain was inserted by thoracic surgeons draining pus and the patient remained ventilated. Over the next 5 days his breathing efforts did not improve significantly, so tracheostomy was performed followed by a repeat CT scan. The scan

| Day No | WBC  | CRP  | Event                                                                 |
|--------|------|------|----------------------------------------------------------------------|
| 1      | 16.3 | 219  | Admission                                                             |
| 4      | 4.73 | 296  | Decreased protein, albumin                                           |
| 7      | 14.75| 214  | Intubation, septic shock                                             |
| 11     | 17.22| 91   | Trans-oral incision                                                  |
| 15     | 12.8 | 17   | Tracheostomy, trans-cervical drainage, debridement and surgical excision of necrotic tissue |
| 19     | 6.9  | 30   | Fifth day of debridement and necrotic tissue removal, wound improved |
| 34     | 5.74 | 115  | Back to the ward                                                     |

Figure 1. CT neck with contrast, axial plane. Retropharyngeal collection.

Figure 2. CT chest, axial plane. Abscess extending into right pleural cavity.

Figure 3. CT neck with contrast after initial intervention draining retropharyngeal abscess. Abscess present in the anterior neck.
showed collection in the anterior tissues of the neck (Figure 3). The patient was then taken back to theatre for trans-cervical drainage of the abscess. Intraoperative findings revealed presence of soft tissues with the appearance of fragmented necrotic “cooked meat”. Bilateral internal jugular veins were inflamed, but at the time of the surgical intervention still patent. Similar appearance of infection was on the strap muscles, sternocleidomastoid muscles on both sides and the thyroid gland. Both sides of the neck were involved from clavicles (in the natural interfacial spaces) levels IB and II under the skull base and reaching till retropharyngeal space. Skin was not involved. Based on the clinical appearance of tissue and intraoperative picture the diagnosis of necrotising fasciitis was made. All the affected facial planes opened and necrotic tissue was removed.

Blood cultures remained negative throughout, initial being performed during the stage of supraglottitis. Neck swabs grew *Pseudomonas aeruginosa*. Microbiological advice was sought and the antibiotic treatment was changed accordingly to clindamycin + meropenem + vancomycin. A day later another CT scan was obtained and revealed locules of gas within the soft tissues as a result of previous surgery. No evidence of mediastinitis was seen. The patient was taken to theatre for daily neck debridement over the course of 10 days. The clinical picture of fasciitis with necrosis progressed for three days, on the 4th day the progression of necrosis stopped. Thereafter six more debridement’s and washouts were performed daily.

Patient was weaned off gradually from the ventilator and inotropic support and was then transferred to the ward after 30 days stay in the intensive care unit.

In the ward he had a slow and prolonged recovery. Initially he was weaned off the tracheostomy and oxygen support and eventually underwent capping and decanulation in conjunction with a speech and language therapist.

Percutaneous enterogastric feeding tube (PEG) was inserted for long-term nutrition and feeding. The patient also underwent extensive physiotherapy in the ward, which was later completed in a local rehabilitation centre. PEG tube was removed 6 months after his discharge from the hospital. Twelve months after his discharge from the hospital patient was discharged from our care without any residual impairment.

**Discussion**

Retro- and parapharyngeal abscess can develop as a complication of tonsillitis, supraglottitis, tuberculosis or other infective conditions in the head and neck. These deep neck space abscess can be caused by aerobic, anaerobic or Gram negative organisms.[4] In our case *Pseudomonas* was grown on culture. Since it can lead to life-threatening emergency with potential airway compromise, it is essential to diagnose it at an early stage.[5] Descending cervical mediastinitis is an uncommonly reported presentation of infection originating in the head or neck and descending into the mediastinum, which is fraught with impressive morbidity and mortality rates of 40% or more.[6] Clinical diagnosis of retropharyngeal abscess or necrotising fasciitis can be challenging in early stages.

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with intrathoracic extension. That case was treated successfully with antibiotics and with a minimally invasive posterior pharyngeal wall incision. Case reports about necrotising fasciitis as a complication of retropharyngeal abscess are rare. Seyhan et al. [2] have described necrotising fasciitis along anterior wall of neck in female with diabetes which later developed into a retropharyngeal collection. We are not aware of any reports of cases of necrotising fasciitis as a complication of supraglottitis together with retropharyngeal collection. We are aware the supraglottitis in our case could have been a secondary phenomenon rather than primary cause of necrotising fasciitis. A recent review of admissions for tonsillitis and neck space abscesses by Lau et al. [16] suggests that there has been a 44% decrease in the tonsillectomy rate from 1991 to 2011 with a 310% increase ($p = .01$) in admissions for tonsillitis and pharyngitis and a 41% increase ($p = .026$) in admissions for retropharyngeal and parapharyngeal abscess.

Therefore, there seems to be an increasing trend which had been reflected in our institution over the past 12 months of retropharyngeal abscess requiring invasive surgical management and hence swift diagnosis and prompt appropriate management is vital.

**Case literature review**

We reviewed various article on Cervical Necrotising Fasciitis (CNF) and found 2627 articles of necrotising fasciitis on Clinical key databases, 408 articles from Pubmed 121 from Medline, 42 from Proquest, 3 from A to Z and 2 from Cochrane search. Our last search completed on 22nd September 2015, when we included cases of CNF leading to mediastinitis and we found out 22 articles (9 with review of literature on CNF) (Table 2).

CNF is a fulminant infection of soft and connective tissues that spreads along the facial planes, inducing posterior venous and arterial thrombosis, followed by necrosis of the skin and other adjacent tissues. It is associated with a high-systemic toxicity and marked compromise of the general state of the patient, with elevated mortality and morbidity rate. It is an uncommon entity that generally presents in the abdomen or limbs, and much more rarely in the cervical region, with published reports of series of cases of no more than 40 patients.[15,17]

Reports of Cervical Necrotising Fasciitis are sporadic, the largest series being 34 cases by Lanisnik et al.[17]

Bahu et al. [18] reviewed the published data from 1973 to 1997 and noted a 35% mortality rate for 70 cases of CNF. Although many case reports and case series are available, no large studies exist, and large series have often mixed CNF data with less-aggressive infection cases. Banerjee et al. [19] and Djupesland [20] reviewed cases of head and neck NF and have also pushed to define CNF as a distinct entity with twice the mortality rate (30% to 38%).

A study from France reviewed 45 patients with CNF. Most of them (78%) were of dental origin; the remaining cases were of pharyngeal origin or had occurred after surgery or trauma.[21] This was confirmed by a report of 11 cases by Wong et al. [22] and a report of 12 cases by Whitesides et al. [23] in which all the cases were odontogenic in origin.

**Conclusions**

- Adult patient presenting with acute tonsillitis without previous episodes of recurrent tonsillitis not improving within days on intravenous antibiotic, then one should think of other diagnosis such as supraglottitis or infectious complication in the neck. In such cases nasopharyngolaryngoscopy should be performed to directly visualise the airway and consider imaging of the neck.
- If there is failure to respond to full medical management early, then CT of the skull base to diaphragm is indicated to rule out the collection in the neck or chest.
- Early diagnosis of retro/para pharyngeal abscess by CT scan and its evacuation can prevent development of necrotising fasciitis.
- Failure to respond to treatment should warrant involvement of a senior clinician early in the process and never delay surgical intervention if required.
- One has to be aware of potential diagnosis of cervical necrotising fasciitis and its seriousness.
- Liaise closely with microbiology for appropriate anti-microbial management.
- In case of developed necrotising fasciitis repeated daily debridement of necrotic tissues in deep neck spaces with application of antiseptic solution to all exposed area is necessary for sufficient local control of infection.

**Disclosure statement**

The study was conducted at University Hospitals Coventry and Warwickshire. The author and co-authors are employed at the same institution. There are no financial interests involved. The article has not been presented before any
| Author                | Year of publication | Number of patients | Mortality rate | Discussion                                                                                                                                                                                                 |
|-----------------------|---------------------|--------------------|----------------|-----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
| Chen et al.           | 2015                | 15                 | 20%            | Advanced techniques for control of the airway have a high rate of success in patients with necrotising fasciitis and could be an appropriate alternative to a traditional airway. Postoperative sedation and analgesia should be considered as routine management of pain and anxiety. |
| Cruz et al.           | 2014                | 6 cases and Review of literature | 0%            | Concerning CNF, early diagnosis and surgical treatment associated to antibiotics and intensive medical care are essential to obtain a favourable outcome.                                                       |
| Jinn-ming-wang et al. | 2013                | 115 cases (4 cases of cervical NF) | 20.9% | Necrotizing fasciitis, although not common, can cause notable rates of morbidity and mortality. It is important to have a high index of suspicion and increase awareness in view of the paucity of specific cutaneous findings early in the course of the disease. Prompt diagnosis and early operative debridement with adequate antibiotics are vital. |
| Thomas et al.         | 2012                | 1 case and Review of literature | 0% | The ability to quickly recognize when additional surgical treatment is necessary might be the key to improving patient survival.                                                                                   |
| Mathew et al.         | 2012                | 1 case and Review of literature | 100% | NF is an uncommon disease that can be extremely devastating. Expedient management of the disease is necessary, because any delay can result in significant morbidity and mortality.                                   |
| Adam et al.           | 2011                | 1 original case and Review of literature | 0% | The case presented in this article provides a good example of the importance of early recognition of NF. Clearly, the need for prompt and aggressive surgical and medical treatment in this immunocompromised patient was shown. |
| Paddy et al.          | 2010                | 1 case and Review of literature | 100% | CNF is a rare and life threatening condition often caused by odontogenic infections, it can initially mimic facial cellulitis due to skin or dental infections. Early recognition and management is essential to improve patient outcome and reduce mortality. |
| Lanisnik et al.       | 2010                | 34                 | 6%             | In this analysis, authors emphasize the importance of an accurate and fast diagnosis, followed by aggressive surgical therapy and a multidisciplinary approach.                                             |
| Karkas et al.         | 2010                | 17                 | 18%            | The aim of this retrospective study was to describe a treatment strategy for CNF with DNM and present a management algorithm for mediastinal extensions of CNF.                                              |
| Zhan et al.           | 2010                | 1                  | 0              | An early diagnosis can be made on the basis of clinical examination, and confirmed by computed tomography (CT) scan, which can prevent complication of NF.                                                          |
| Guido et al.          | 2010                | 1                  | 0              | The aim of study is to start early treatment to limit extension of the infection by early diagnosis and appropriate fast therapy (medical management and surgical debridement). It is also very important to eliminate the odontogenic source: caries, endodontic and periodontal abscesses. |
| Faisal et al.         | 2009                | 1 original case and Review of literature | 100% | Prompt identification of the patient at risk for this disease process as well as aggressive management is critical. What remains is a methodology to identify the presence of CNF versus cellulitis or abscess. |
| Giovanna et al.       | 2007                | 1                  | 100%           | The complete surgical exploration and extensive drainage with appropriate medical management could have reduced the morbidity and mortality rates for cervical necrotizing fasciitis and could have prevented fatal illness like descending mediastinitis. |
| Mora et al.           | 2004                | 21                 | 25-40%         | The aim of this is to help define the clinical criteria and diagnostic procedures that will improve the early diagnosis of mediastinal sepsis secondary to neck fasciitis and to suggest optimal treatment approaches. |
| Umeda et al.          | 2003                | 9 original cases, 125 cases- review of literature | 19.2% | The aims of this study were to describe the condition of this rare disease NF and to find factors affecting the mortality                                                                                           |
| Bahu et al.           | 2001                | 10                 | 10%            | Patients were analysed for source and extent of infection, microbiology, co-morbidities, antimicrobial therapy, hospital days, surgical interventions, complications, and outcomes. |
| Lee Whiteside et al.  | 2000                | 12 cases and Review of literature | 0% | Early surgical intervention and the use of Hyperbaric oxygen therapy decreases morbidity and improves the clinical outcome.                                                                                     |

(continued)
professional otolaryngological association and is an original work.

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