LUDWIG'S ANGINA AND PARA-PNEUMONIC EFFUSION IN A CHILD: CASE REPORT

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ABSTRACT

INTRODUCTION: Ludwig's angina as a severe odontogenic infection and para-pneumonic effusions are two distinct clinical entities with their unique potential challenges, spanning various specialties. Ludwig's angina is an uncommon presentation among children and could be an indication of child neglect. The spread of severe odontogenic infections has been found to cause complications such as parapneumonic effusion, mediastinitis, intracranial abscesses, thrombophlebitis of the internal jugular vein, empyema, pericardial effusion, osteomyelitis of the mandible, subphrenic abscess, and aspiration pneumonia, among others. The management of both Ludwig's angina and para-pneumonic effusion requires early aggressive medical and surgical intervention for patient survival.

CASE REPORT: In this case report, we discuss a five-year-old boy who presented with Ludwig's angina, in whom para-pneumonic effusion was found. He was successfully managed with a multidisciplinary approach. We also reviewed relevant clinical anatomy and the etiopathogenesis of Ludwig's angina as a cause of para-pneumonic effusion, as well as the the diagnostic sequence of both clinical entities.

CONCLUSION: Cervicofacial odontogenic infections can be complicated by para-pneumonic effusion, which could be fatal. Early recognition and initiation of treatment remain key to preventing mortality. Airway observation and management, aggressive antimicrobial therapy, and surgical decompression with removal of the source of infection remain vital in the successful management of this condition. In situations where the patient is medically compromised or develops a medical complication, a multidisciplinary approach is necessary for patient management.

KEYWORDS: Ludwig's angina, parapneumonic effusion, severe odontogenic infection

INTRODUCTION

Ludwig's angina (LA) is a potentially life-threatening and rapidly progressive bilateral cellulitis of the floor of the mouth (sublingual space) and neck (submental and submandibular spaces) with induration [1–3] Ludwig's angina is more prevalent in middle-aged individuals, but about a third of paediatric cases have been reported with its current incidence about 24-30% [4–6]. LA is observed to be a fatal disease because it can cause airway embarrassment that can lead to death if not managed timely[6–8]. LA in children is more severe and deadly compared to adults due to the rapid progression of the disease[9].

In general, odontogenic sources of infection have been observed to account for the majority of LA, and in children, it accounts for about 50% of cases[5,9]. Other causes and predisposing factors for LA in children include oral viral infections, upper respiratory tract infections, penetrating injuries to the tongue and the floor of the mouth, otitis media, mandibular fractures, mucosal lacerations, sialadenitis of the submandibular gland, diabetes mellitus, and an immunocompromised state[4,5]. On the contrary, about 25% of LA reported among the paediatric population had no associated cause or risk factors[5]. Airway obstruction is a common complication among patients with LA and is more likely to present in children due to their relatively higher-positioned larynx compared to adults.[10] Airway management plays a major role in the treatment of patients with LA. Other reported complications of LA include pleural effusion, carotid arterial rupture or sheath abscess, thrombophlebitis of the internal jugular vein, mediastinitis, empyema, pericardial effusion, osteomyelitis of the mandible, subphrenic abscess and aspiration pneumonia[7,11,12]. We report on the management of a case of a five-year-old boy with Ludwig's angina, complicated by para-pneumonic effusion.

Verbal consent was sought from the mother of the patient and permission was given to use patient information and pictures for case report.

CASE PRESENTATION

A five-year-old boy presented to the maxillofacial unit of Korle Bu Teaching Hospital (KBTH) with a one-week history of severe toothache and an associated four-day history of progressive submandibular swelling, which started as a right submandibular nodule. Patient at presentation had symptoms of fever, odynophagia, and dysphagia to solids. Topical herbal medication and over-the-counter medications were used to manage the child's condition at home, but symptoms worsened over time, thus reporting to our facility. He had no known underlying medical condition.

On arrival, the child was conscious but lethargic and showed signs suggestive of respiratory distress (stridor and dyspnoea). Further examination revealed cervicofacial swelling that was firm, diffuse, tender, and bilateral involving the submandibular, sub-masseteric spaces, and submental space. There was trismus (inter incisal distance was 1cm wide), elevated tongue, purulent discharge from the right buccal sulcus, extensive caries of all first and second molar teeth in the maxilla and mandible, and he was mildly dehydrated. Chest examination revealed reduced air entry in the middle and lower zones bilaterally, having broncho vesicular breath sounds were transmitted with coarse crepitation in all lung zones. His vitals at presentation were; respiratory rate 52cpm, oxygen saturation of 92%-95% (room air), heart rate 148bpm, and temperature of 38 degrees Celsius. Child's weight was 17kg which was normal for his age. Complete blood count revealed low haemoglobin level of

7.5g/dl, elevated white cell count of 28.81 x 109/L with differential neutrophilia 26.02 x 109/L and low platelet count of 31 x 109/L and retroviral screen was non-reactive. Considering the clinical presentation and laboratory results, a diagnosis of Ludwig's angina with moderate anaemia was made. Patient was admitted and medical treatment initiated with IV amoxiclav 600mg, IV metronidazole 150mg, IV paracetamol 500mg, IV dexamethasone 2mg, IVF 1/5 normal saline in 4.3% dextrose at 45mls/hr and oxygen administered via nasal prongs. An emergency tracheostomy, incision and drainage (decompression) as well as extraction of all grossly carious first and second molar teeth were done under General Anaesthesia (Figure 1). Patient was transfused with 350mls of whole blood and 2units of FFPs in theatre. Samples for culture and sensitivity (aspirate and blood) were taken intraoperatively, but results showed no bacterial culture growth.

Figure 1: Incision and drainage in the bilateral submandibular, submental, and right sub-masseteric regions done under General Anaesthesia.

Post-operatively (post-op day 1), the patient recorded an average SpO2 of 83% and was coughing intermittently whilst on supplementary oxygen of 4L/min delivered via tracheostomy mask. Chest examination revealed: stony dullness to percussion in the left and right mid and lower zones, diminished chest expansion on the left, decreased breath sounds, tactile vocal fremitus was decreased. A plain chest x-ray (figure 2) revealed opacifications in all the lung zones; a collapsed right lung consistent with para-pneumonic effusion with pneumothorax of the right lung.

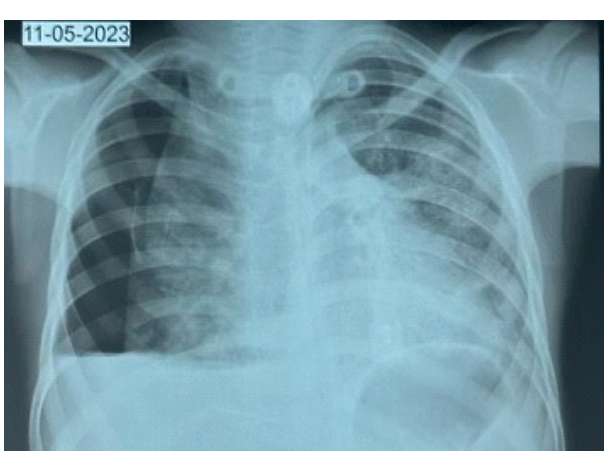

Figure 2: A plain chest x-ray showing opacification of all lung zones and collapsed right lung.

A diagnosis of para-pneumonic effusion with pneumothorax complicating Ludwig's angina was made on Cardiothoracic team evaluation. This was managed by right thoracocentesis by the cardiothoracic unit team (figure 3). Upon insertion of the chest tube, there was initial gushing out of air evidenced by bubbling in the underwater seal, this was followed by drainage of 50mls of serous fluid. The pleural aspirate was obtained for culture and sensitivity, yielding no bacterial growth. Medications were reviewed with the addition of IV Rocephin 1g to previous medication. The patient was closely monitored during admission and co-managed by the Paediatricians, Cardiothoracic unit team, Physiotherapists, Dietitians, and the Otorhinolaryngology team. Serial chest radiographs and laboratory investigations were requested to monitor the patient's progress.

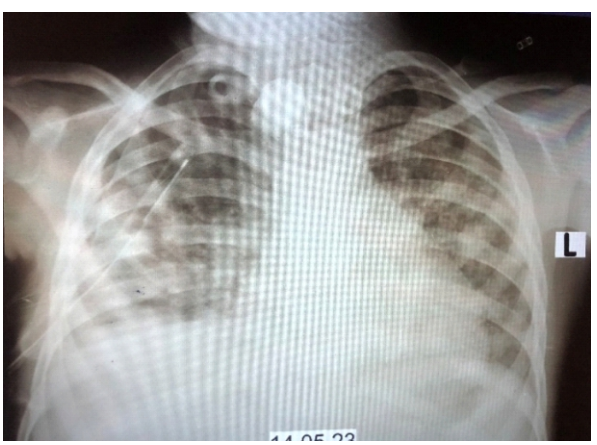

Figure 3: A plain chest x-ray showing tracheostomy tube and chest tube in situ and evidence of collapsed right lung, generalized opacification of all lung zones.

The patient recovered fully and was discharged after 4 weeks of admission. The patient had corrugated drains, chest tube, and tracheotomy tube removed prior to discharge. A panoramic radiograph was requested and child was referred to the paedodontist for further assessment and management.

Figure 4: A clinically well-looking boy with no obvious facial swelling prior to discharge.

DISCUSSION

The rare progression of Ludwig's angina (LA) and the resultant thoracic empyema noted in this child patient warrants analysis. LA is deep fascial space infection of the cervical region and is a common form of presentation of spreading odontogenic infection with incidence rate of 52% [13]. The odontogenic sources of infection have been observed to be the major cause of LA in both paediatric (50%) and adult (70% - 90%) population[5,14]. Similarly, our patient presented with an underlying dental abscess from the carious mandibular tooth. Another common cause of LA identified among the paediatric population is upper respiratory tract infections (URTI) [4]

Ludwig's angina of odontogenic origin is mostly initiated by caries and progresses to the apical region of the tooth or periodontal disease. The infection then enters the medullary bone, forms pus, and perforates the cortical bone, where it appears clinically as a swelling [5]. Failure to manage infection appropriately at this stage results in the spread along the facial planes associated with complications that may be fatal[15]. Spreading odontogenic infections within the head and neck are predicted by the attachment of the cervical fascial layers to neck structures[16,17]. In cases of LA, which result from a mandibular source of infection, the primary potential spaces involved are the submandibular, sublingual, and submental spaces bilaterally, as seen in our patient. 9,18 Occasionally, the spread can involve other adjacent spaces, such as the sub-masseteric, pterygomandibular, buccal, and vestibular spaces[18]. LA can also spread to involve the parapharyngeal, retropharyngeal, pre-tracheal spaces, and mediastinum[16,18]. As observed in this patient, the spread of infection involved the deep spaces of the neck and also within the thorax, resulting in a para-pneumonic effusion. Thoracic empyema/ para-pneumonic effusion is a relatively uncommon complication of LA[12]. The three (3) identified routes of spread of neck infection to the thorax are the retropharyngeal, pre-tracheal, and vascular spaces of the neck[12]. These above spaces represent secondary spaces of spread in mandibular infection in patients with LA[12]. The main factors that facilitate the spread of odontogenic infection from these spaces include the patient's immunocompromised state and the virulence of microorganisms, which is based on the fact that toxins produced by these virulent organisms cause destruction of connective tissue in these spaces, creating a conduit for the spread of infection[9]. In this patient, the systemic condition identified at presentation was moderate anaemia, which could be an indication of malnutrition[19].

Nevertheless, the typical systemic conditions like HIV/AIDS, uncontrolled diabetes, as well as therapeutic agents such as chemotherapy drugs and long-term corticosteroid use that are usually associated with an immune suppressive state were absent[12]. In a study by Fomete et al., anaemia caused by nutritional deficiency was found in one patient managed for empyema thoracis resulting from a spreading odontogenic infection, which was similar to that seen in our patient[19]. Other host factors mentioned in this study were diabetes mellitus and psychiatric disorders[19].

Severe odontogenic infections, such as Ludwig's angina, are considered polymicrobial in origin and encompass a range of microorganisms, including both Gram-positive and Gram-negative bacteria, as well as aerobic andnaerobic microorganisms, hence the response to broad-spectrum antibiotics[20,21]. The commonest organisms are Streptococcus, Staphylococcus, Fusobacterium, Klebsiella, Bacteroides, and Actinomyces[9,22]. Some of these organisms are noted to be highly virulent due to their ability to produce toxins, including hyaluronidase, collagenase, and streptokinase, which destroy connective tissues.

Para-pneumonic effusion (empyema thoracis) develops gradually from contiguous infection of mediastinum and adjacent structures, resulting in pleural inflammation with subsequent increased pleural permeability, leakage of proteins, fluid, and leucocytes into the pleural space[23]. Progress of the infection due to inadequate treatment can result in restrictive lung disease as a result of fibroblast transformation of fibrin membranes (produced in the fibrinopurulent stage) in pleural space into non-elastic pleural peel[23]

In the paediatric population, para-pneumonic effusions are seen in about 2-12% of children with pneumonia mostly caused by Streptococcus species, and about 28% of these require hospitalization[24]. Other aerobic organisms, such as Staphylococcus aureus, and anaerobic infections secondary to aspiration and mycobacterial infections in immunocompromised patients have also been reported[25]. These microorganisms commonly seen in paediatric empyema thoracis are also commonly isolated and cultured for LA. The occurrence of a para-pneumonic effusion in this child patient with LA could be due to the natural progression of the infectious process, given the similarities in causative agents. However, the cultures of both pus aspirate and thoracocentesis yielded no bacterial growth. The absence of bacterial growth in the samples obtained from our patient may be due to the patient's previous use of antibiotics or to laboratory errors resulting from either poor sample-taking technique (timing) or inadequate transportation and processing of the sample. The cause of the para-pneumonic effusion in this patient can also be attributed to the aspiration of intraoral pus discharge. Bacterial cultures, either via swab or needle aspiration, had limited value in the diagnosis of LA and para-pneumonic effusion in the case presentation due to no bacterial growth; however, it is still necessary to obtain samples. Blood cultures, however, must be obtained to determine hematogenous spread. [26] Clinical evaluation is a major diagnostic factor for Ludwig's angina. [19] Although imaging may help, it does not play any direct role in the initial assessment of the patient. It is, therefore, essential to ensure that airway patency is maintained and the risk of airway obstruction is minimized or the airway has been secured by intubation or tracheostomy before sending the patient for the required imaging[3,6,20]. There was no imaging for the patient, as stated in the case report, prior to surgical drainage of the abscess because he needed an immediate emergency tracheostomy to secure the airway. Imaging that can be taken to assess the severity of Ludwig's angina is a CT scan of the neck as well as an ultrasound of the cervical region[5] . Ludwig's angina may be complicated by sepsis, pneumonia, asphyxia, empyema, pericarditis, and mediastinitis[7]. The current mortality rate in the paediatric population is 10%-17%. [24,25]

Diagnosis of para-pneumonic effusion is based on clinical, radiographic as well as laboratory findings[23,25]. Clinical manifestations include severe productive cough, fever, dyspnoea, and chest pain.[19,23] . Physical examination findings may vary depending on duration of the illness and the organisms involved. This includes dullness to percussion and decreased breath sounds, as well as crackles or crepitations and reduced air entry on

auscultation. [19,23,25] Laboratory studies include a complete blood count and a total protein level (as part of the liver function test)[19].

Radiographic evaluation is a primary tool for diagnosing and evaluating the progression of para-pneumonic effusion. A standard plain chest radiograph is the first step to assess for pleural fluid. Serial chest radiographs are then taken to evaluate the progression/ resolution of the illness. Ultrasonography, computerized tomography, and magnetic resonance imaging have been outlined to be relevant in treatment planning[19,23].

Aggressive treatment with surgical decompression to preserve the oropharyngeal airway and reduce mortality is still the first choice of treatment in Ghana[13]. The patient's condition in the case report was complicated by para-pneumonic effusion with associated pneumothorax, for which a broader spectrum of antibiotics was introduced, as well as thoracocentesis done for the right lung effusion. Empirical antibiotics, which comprise penicillin, metronidazole, and gentamycin (when not contraindicated), are considered the first-line antibiotics for treating spreading orofacial infections[2,3,13,19,27,28]. In our patient, Rocephin was added to penicillin and metronidazole to manage his condition successfully. The addition of Rocephin (ceftriaxone) to these empirical antibiotics may have masked their efficacy in managing such conditions[19].In a Nigerian study by Fomete et al.[19], these empirical antibiotics were used to effectively manage cases of spreading odontogenic infections complicated by parapneumonic effusion. Management of parapneumonic effusions focuses on controlling infection, which involves draining pleural fluid, using broad-spectrum antibiotics or antibiotics based on cultured bacteria and providing supplemental oxygen depending on the patient's oxygen saturation[23,25]. The lack of bacterial growth in this patient resulted in the use of broad-spectrum antibiotics instead of targeted therapy. In this case report, the patient was successfully managed using a similar treatment with the addition of nutritional support and incentive spirometry[23].

A multidisciplinary approach to managing the complication from Ludwig's angina in children will include an oral and maxillofacial surgeon, cardiothoracic surgeon, otorhinolaryngologist, anaesthetist, and paediatrician.

LIMITATION

- The inability to do imaging of the head and neck region to identify the anatomic spaces involved in the orofacial infection
- The absence of bacterial growth in pus aspirate from the cervical region and pleural aspirate was a challenge in diagnosing thoracic empyema as a sequela of Ludwig's angina, and it also presented a treatment challenge, as empirical antibiotics were used instead of targeted therapy.

CONCLUSION

Ludwig's angina remains a potentially life-threatening infection among the paediatric population, which can be complicated by mediastinitis. Early recognition and initiation of treatment remain key to preventing mortality. Airway observation and management, aggressive antimicrobial therapy, and surgical decompression with removal of the source of infection remain vital in the successful management of this condition. In situations where the patient is medically compromised or develops a medical complication, a multidisciplinary approach is necessary for patient management.

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