

Case Report

Diagnosis and treatment of acute descending necrotising mediastinitis caused by pediatric oropharyngeal trauma

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Abstract: Acute descending necrotizing mediastinitis (ADNM) is an acute necrotizing infection in which oropharyngeal infection descends and involves the mediastinum, causing respiratory distress and sepsis. This paper reports the successful remedy of an ADNM case caused by oropharyngeal trauma in a 2-year-old child. Timely treatment, including abscess drainage in the chest, adequate application of broad-spectrum antibiotics and fully symptomatic support treatment are important steps towards healing. In this report, for suspected of oesophageal perforation, feeding with duodenal intubation was performed. Admitting physicians could use this report to familiarize themselves with the handling necessary for this type of rare but deadly emergency case.

Keywords: Acute necrotizing mediastinitis, oesophageal perforation, paediatric, trauma

Introduction

The occurrence of paediatric acute descending necrotising mediastinitis (ADNM) is relatively rare because of the extensive use of antibiotics and the effective control of upper respiratory tract infection. Since its first report in 1938, ADNM has been given considerable attention by professional physicians because of its low incidence but high risk. Reports on ADNM caused by paediatric oropharyngeal trauma are relatively rare. In this paper, a successful remedy for a case of ADNM in a 2-year-old child caused by oropharyngeal trauma was analysed and discussed. This case study may serve as a reference for the diagnosis and treatment information of ADNM for otolaryngologists and paediatric emergency physicians.

Case report

The patient was a 2-year-old male who fell on November 28, 2012 with chopsticks in his mouth. This study was conducted in accordance with the declaration of Helsinki. This study was conducted with approval from the Ethics

Committee of Shanghai Jiao Tong University. Written informed consent was obtained from all participants' guardians. He was sent to a local hospital for medical treatment of mouth bleeding. The oropharyngeal examination did not exhibit any obvious abnormality, so he was discharged and follow-up treatment was performed at home. However, his jaw swelled during the night. The next morning, the parents found that the boy's mandibular swelling worsened. Thus, he was brought to our hospital for physical examination, which revealed that the boy's mandibular swelling was significant, with tenderness. Skin temperature was normal, but mucosal damage can be observed on the right side of the sublingual mouth floor, without any significant leakage, bleeding or swelling. The child's spirit was poor, his vital signs were stable and he was not suffering from fever. The parents revealed that the boy's eating and drinking conditions were normal, without vomiting, dysphagia, shortness of breath or discomfort. The emergency blood routine test indicated the following data: WBC $18.6 \times 10^9/L$ and C-reactive protein (CRP) $>160 \text{ mg/L}$. Cefuroxime (100 mg/kg/day, intravenous infusion, Bid),

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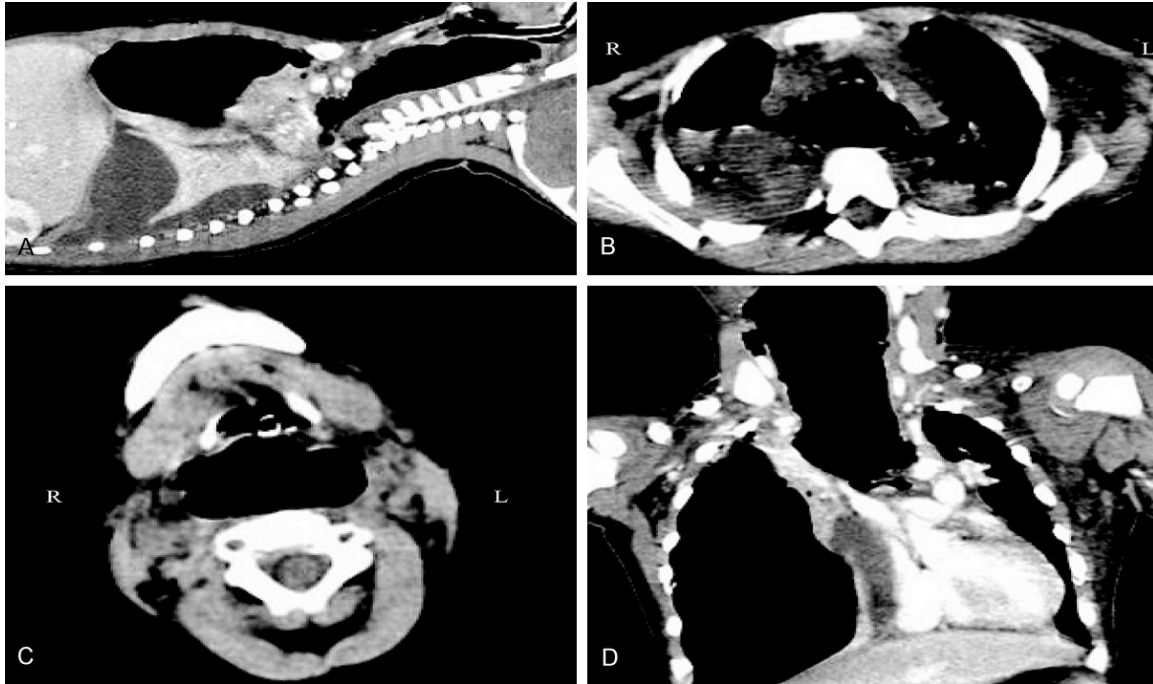


Figure 1. Enhanced neck-chest computed tomography (CT) scanning before treatment. A: Large gas accumulation is observed in the pre-vertebral space in median sagittal section of neck CT scanning. B: Large gas accumulation is observed in the pre-vertebral space in transverse section of chest CT scanning accompanied by corresponding hydropneumothorax and atelectasis changes in the right lung. C: Large gas accumulation is observed in the pre-vertebral section in transverse section of neck CT scanning. D: Subcutaneous pneumatosis in the mediastinum, throat gap, and bilateral chest is noted. Left pleural effusion is apparent in the coronary section of neck-chest CT scanning.

ornidazole (100 ml/day, intravenous injection, qd) and dexamethasone injection (5 mg/day, intravenous infusion, qd) were used. The patient was admitted for further observation.

On the second day, the boy's swollen jaw significantly decreased, without fever or particular discomfort. The parents requested that the boy be taken home, but the chief physician stated that the disease situation was not stable. Potential risk of breathing difficulties existed, so the patient was admitted into the emergency department. At noon, the patient suddenly exhibited shortness of breath and his neck swelled, so he was administered oxygen inhalation and subjected to electrocardiography. Simultaneously, emergency bedside endotracheal intubation was performed with the assistance of the Department of Anaesthesia. However, the procedure failed because of unclear glottic exposure and blood decrease in oxygen saturation. Thus, the patient was immediately transferred to the intensive care unit (ICU). After admission to the Intensive Care Unit (ICU), the patient exhibited subcutaneous emphyse-

ma in the neck and upper chest, with obvious crepitus. Dyspnoea sharply increased, and repeated intubation failed because the pharyngeal cavity was narrow and the glottis could not be exposed. Pernasal intubation was selected and finally succeeded with the guidance of the fibrolaryngoscope. Emergency chest radiography indicated that subcutaneous pneumatosis existed in the bilateral neck and the right armpit. Minor inflammation existed in the bilateral lungs, and the left lower part exhibited partial atelectasis. Enhanced neck-chest computed tomography (CT) scanning indicated that a large area of gas accumulated in the pre-vertebral space and that the right chest had hydropneumothorax, accompanied by changes in right lung atelectasis and mediastinum; subcutaneous pneumatosis appeared in the throat clearance and bilateral chest, and effusion appeared in the left thoracic cavity (**Figure 1**). The patient immediately underwent subcutaneous puncture for evacuating, and closed-type drainage was performed towards the right thoracic cavity. The drainage liquid was yellow and turbid,

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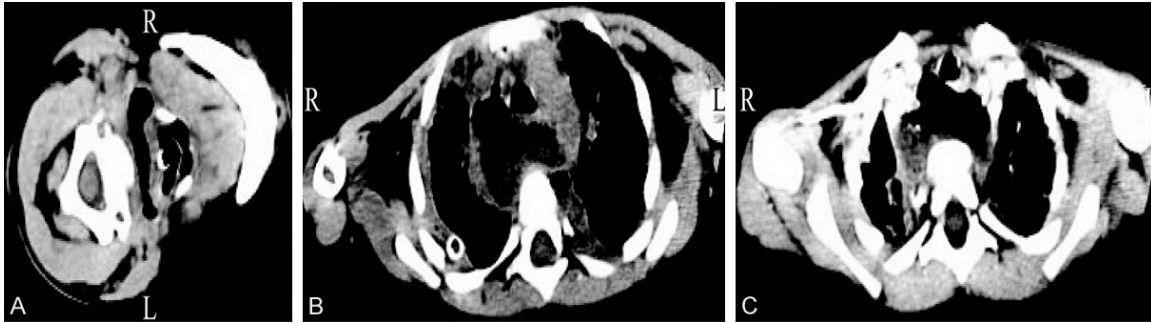


Figure 2. Enhanced neck-chest computed tomography (CT) scanning after one month treatment. A: Abnormal mediastinal sac shadow is observed in the retropharyngeal and right parapharyngeal space, accompanied by gas and contents. B: The bilateral lungs are extensively infected. C: The right pneumothorax and effusion are significantly reduced when compared to previous condition.

with negative bacterial cultivation. Although no positive pathogens were detected, combined with the symptoms and signs, the patient was determined to have suffered from deep neck space infection, which moved downward and involved the mediastinum. These data met the diagnosis of DNM; therefore, the patient was given the broad-spectrum antibiotics Mepem (10 mg/kg/time, intravenous infusion, q8h), Vancocin (10 mg/kg/time, intravenous infusion, q8h) for anti-infection, and methylprednisolone (5 mg/kg/day, intravenous infusion, bid) for anti-inflammatory treatment based on experience. At the same time, the patient was nasally fed with the strengthened supportive treatment.

At 1 month after the treatment, the vital signs of the patient became stable, and the chest drainage was gradually reduced. However, clear fluid drainage was still leaking out daily. The CT scan review indicated abnormal sac shadow in the mediastinum behind the retropharynx and right parapharyngeal space, accompanied by gas and contents; the bilateral lungs suffered from extensive infection, whereas the right pneumothorax and effusion were significantly reduced compared with the previous analysis (**Figure 2**). Analysis of the disease suggested that the lung condition was not improved, mediastinal abscess formed and oesophageal damages may exist. In addition, the nasogastric nutrition tube may lead to persistent infection in the perforation site and spread to the mediastinum. The mediastinal fluid volume was too large to be absorbed, thereby affecting the healing process. After the combined consultation with the Department of Cardiac Surgery, the surgeons performed thoracoscope-assisted left mediastinal incision for the drainage

and changed the gastric tube nutrition to the duodenal feeding under the guidance of an endoscope. A large amount of purulent secretions was drained from the mediastinum. The CT scan suggested significant improvement in the lungs after 10 days (**Figure 3**); hence, the ventilator was adjusted to the artificial nose oxygen inhalation. After 2 days of observation, the patient exhibited better spontaneous breathing, with 99% oxygen saturation and without significant liquid drainage from the bilateral drainage tubes. In addition, the patient had smooth breathing after removing the tracheal cannula and drainage tube. The duodenal feeding was continued for 10 days until the oral intake of milk exhibited no obvious abnormalities, and then the duodenal feeding tube was removed. After 1 week of observation, the CT scan review suggested that the abnormal sac shadow in the neck and upper mediastinum shrank, the right pneumothorax was fundamentally absorbed and the pulmonary inflammation improved (**Figure 4**). The continuous observation indicated that the vital signs of the patient were stable; thus, the patient was discharged, with a total hospitalisation period of 66 days.

The follow-up continued for 3 months. The follow up date indicated that the normal daily activities of the patient were normal. No abruptness, swallowing difficulty and breathing difficulty were observed after any activity. Moreover, the patient's weight increased steadily, and the various indicators of growth and development were normal.

Discussion

ADNM is a necrotising fasciitis or necrotising cellulitis that starts from the oropharynx or

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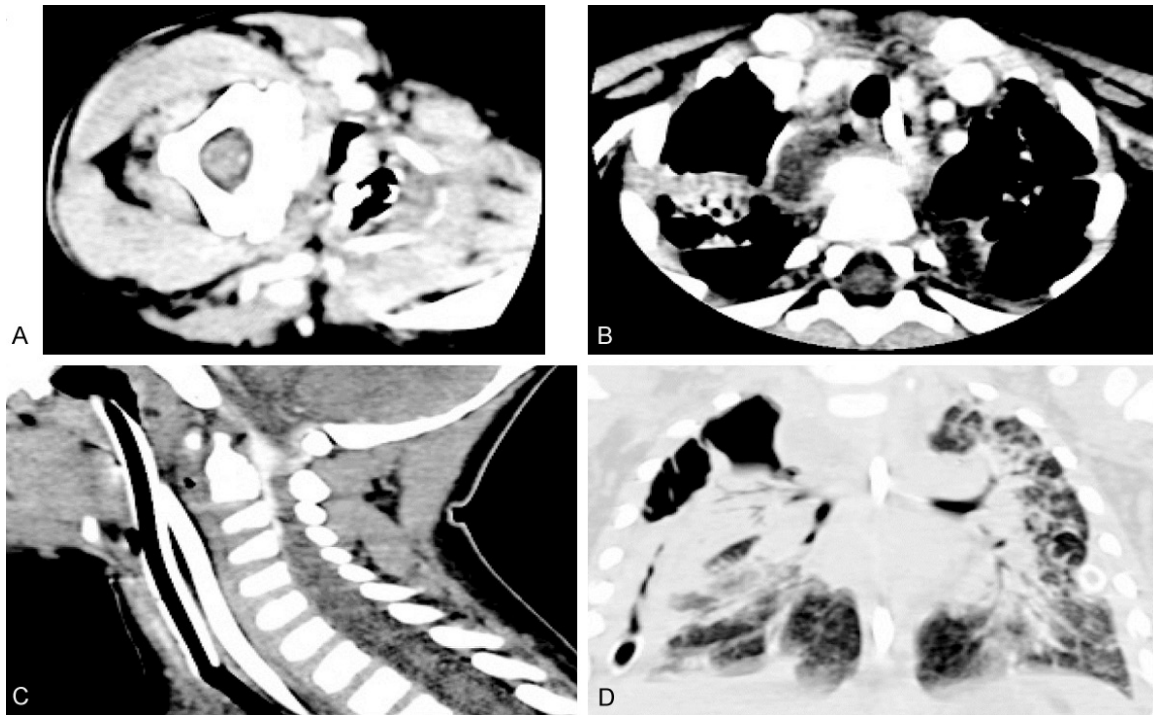


Figure 3. Enhanced neck-chest computed tomography (CT) scanning 10 days after the left mediastinal incision for the drainage and the duodenal feeding. A: The sac shadow in the retropharyngeal space decreased significantly in the coronary section of neck CT scanning. B: The gas accumulation is reduced in the pre-vertebral space in transverse section of chest CT scanning and the lung condition has improved. C: The gas accumulation in the pre-vertebral space nearly disappeared in coronary section of neck CT scanning. D: The bilateral lungs are infected but the effusion is reduced.

head and neck, and then descends to the mediastinum through the deep cervical fascia. ADNMs was first reported in 1938 by Pearse [1], who suggested that the infection spreads through three paths, namely, the retropharyngeal space, the perivascular fascial space, and the anterior trachea space. ADNMs is characterised by the absence of obstacles among the spaces, the formation of negative pressure in the chest during breathing, and the presence of tissue necrosis and gas-generating biotical creatures. Although the occurrence of ADNMs is rare, it often causes respiratory distress and organ failure because its infection could spread rapidly in a short period. ADNMs may sometimes cause sepsis, which accounts for its high mortality rate [2-4]. Before 1990, the mortality of DNM has been reported to range from 40% to 50%. However, after 1990, the highest mortality rate reported was only 11% because of the application of advanced surgical interventions [5].

The common causes of ADNMs are retropharyngeal abscess, parapharyngeal abscess, peritonsillar abscess, odontogenic abscess, epi-

glottitis, deep cervical lymphadenitis, mumps, thyroiditis, trauma, traumatic pharyngeal, oesophageal foreign materials and infection, endoscopy and other iatrogenic causes [6]. ADNMs is more common in children within 0 to 3 years old and relatively rare in adults. Previous literature (1990 to 1996) [7] reported that ADNMs is mainly caused by odontogenic reasons. By contrast, recent literature has shown [5] that ADNMs is mainly due to throat infection, followed by throat injuries caused by foreign materials, iatrogenic perforation and neck infections. The trauma-caused ADNMs in children is rare; it generally occurs in situations where the child has some objects (such as toy, chopstick, pencil or toothbrush) in the mouth when running and falls down [8-11]. Several DNM cases caused by foreign material-induced trauma have been reported, most which were caused by retropharyngeal space infections that resulted in throat wall injuries by sharp foreign materials [12, 13]. Some cases in which the ingestion of foreign materials injured the oesophageal tract and then caused mediastinitis have also been reported [14]. In 2006, Schulz et al. [15] reported one case in which an

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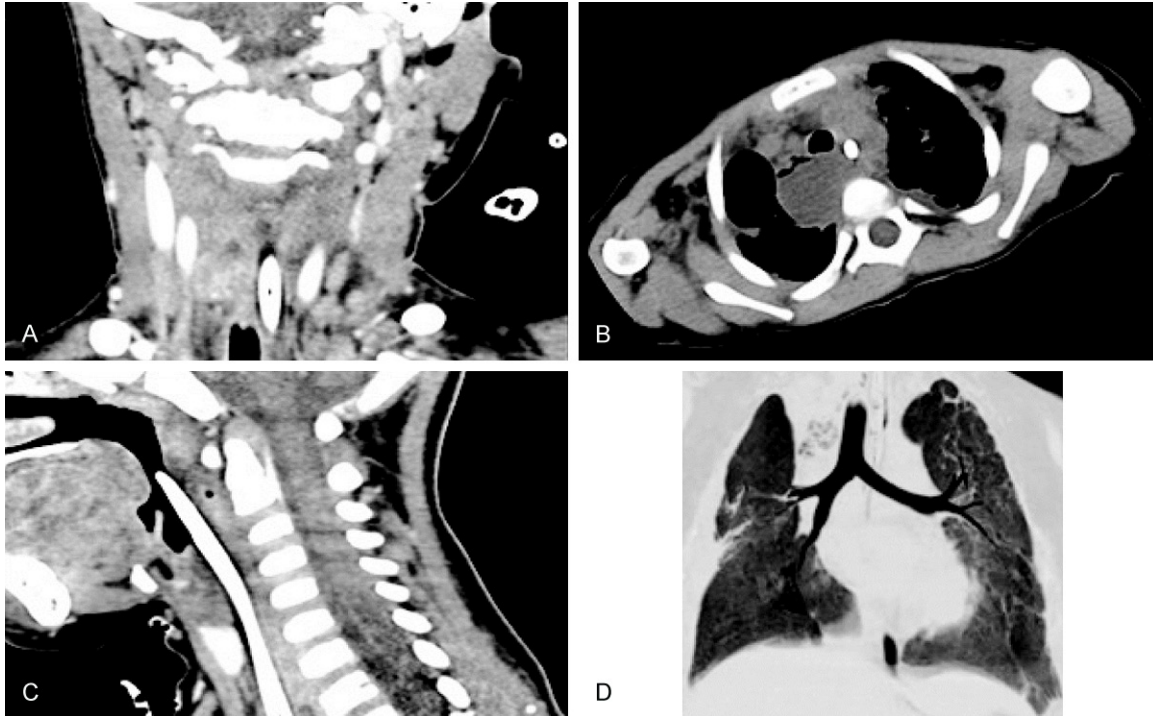


Figure 4. Enhanced neck-chest computed tomography (CT) scanning 2 months after treatment. A: The abnormal sac shadow in the neck shrank. B: The abnormal sac shadow in the upper mediastinum shrank. C: The sac shadow in the retropharyngeal space disappeared. D: The right pneumothorax is fundamentally absorbed, and lung inflammation is reduced.

8-year-old girl suffering from cerebral palsy mistakenly ingested some foreign material that caused oesophageal perforation and eventually DNM. The girl died during the treatment because it was not found timely. The characteristic of such case is oesophageal damage. Once suspected, the foreign material needs to be removed immediately. In addition, the patient should be given immediate attention to prevent the sustained repeated infections in the oesophagus damage by food, which would cause mediastinitis. In our case, the child fell down with chopsticks in his mouth, so it was a DNM case caused by trauma. Given that the wound in the mouth floor was small and closed quickly, the parents thought that the disease was not heavy, and the local hospital did not make any treatment. When admitted into our hospital, the case was given timely attention, examination and treatment; thus, the patient ultimately recovered. The diagnosis and treatment of trauma in the oropharynx, head and neck should not only be limited to the condition of the focal wound. Deep tissue infections caused by small trauma should also be considered because such infections could spread and lead to DNM.

The clinical manifestations of DNM are acute, often accompanied with chills, fever, sore throat, swallowing difficulty, vague speech, neck swelling, chest pain, limited neck mobility and breathing difficulty. The disease progression is fast, and the complications are complex. The early symptoms of DNM are not typical; thus, the probability of missed and wrong diagnosis is high. Early diagnosis and treatment of DNM are necessary. The disease history of the suspected patient should be inquired carefully, and enhanced neck and chest CT scanning should be performed as early as possible. In addition, condition changes should be given close observation, and intervention should be carried out immediately when the patient's condition changes. In 1983, Estrera et al. [16] defined the DNM diagnostic points in detail as follows: 1) clinical manifestations of oropharyngeal infection (odontogenic, periamygdalitis, retropharyngeal abscess or throat infection after trauma); 2) X-rays revealing the characteristic performance of mediastinitis; 3) surgical operation or autopsy providing evidence of mediastinitis; and 4) certain connection between the oropharyngeal infection and mediastinum. CT examination has an important role

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in the diagnosis. Estrera et al. [16] first highlighted the diagnostic value of CT examination in DNM cases with nonspecific symptoms. CT examination can detect the retropharyngeal space and mediastinal pneumatosis or effusion in many cases with nonspecific symptoms. Hence, CT can facilitate early intervention of the disease and greatly improve the cure rate. In the present case, the patient was admitted into our hospital a day after the trauma. Considering that oropharyngeal trauma may be followed by secondary deep space infection and critical condition, the patient was hospitalised immediately. This situation reflects that our hospital is fully aware of DNM and its diagnosis. Upon admission, the emergency neck-chest CT scanning revealed the typical manifestations of pneumatosis and effusion in the retropharyngeal space, mediastinum and thoracic cavity, supporting the diagnosis of DNM; hence, treatment was conducted immediately. CT examination had an important function from the early diagnosis to the judgment of treatment effects, providing the exact basis to determine the disease condition and treatment effects. Combined with the previous reports and this case, we conclude that early diagnosis and treatment through CT examination are necessary to improve the cure rate of DNM.

DNM infections are mostly caused by the combination of aerobic and anaerobic bacteria. In an earlier study, Ridder et al. [5] reported that the most common aerobic and anaerobic bacteria causing DNM infections are *Streptococcus* and *Bacteroides* spp., respectively. Recent studies [17, 18] have shown that DNM is mainly caused by the mixed infection of high-virulent anaerobic and aerobic bacteria. Some aerogenic bacteria isolated from head-neck infections include *Clostridium* [19], *Bacteroides*, and *Fusobacterium* [20]. Meanwhile, Olushola et al. was able to isolate the aerobic bacterium *Klebsiella pneumoniae*. Positive results are difficult to obtain during cultivation. Karkas et al. [21] proposed that deep cervical fascia infections have two clinical forms: purulent and pneumatosis. The former is characterised by large formation of effusion, whereas the latter is characterised by large accumulation of gas. However, purulent infection is more common clinically. CT manifestation of this infection includes effusion occupation in the retropharyngeal space and mediastinum. Thus,

purulent infection could be easily diagnosed as retropharyngeal abscess and mediastinitis. Afolabi et al. [14] reported a rare case of fish-bone-damage-caused pharyngeal abscess mixed with aerogen infection. X-ray scans revealed abundant gas accumulation in the widened retropharyngeal space; such accumulation can easily cause misdiagnosis. **In the early and middle courses of the disease in the present case, the clinical manifestation was the accumulation of gas between the neck and chest gaps.** The cultivation result of the drained fluid was negative. The use of broad-spectrum antibiotics in the early stage might have affected the cultivation results. Although several cultivations did not provide any evidence of aerogen infection, the clinical manifestation can help to infer the possible aerogen infection in the deep neck and mediastinum. When confronting cases that first appeared as emphysema, clinicians should have this awareness to avoid misdiagnosis.

Decades of clinical experience have confirmed that the treatment principles of DNM are to maintain the airway unobstructed, adequate application of broad-spectrum antibiotics and timely surgical drainage (including the drainage in the neck, chest and mediastinum) [22-24]. 1) In the present case, intubation was performed immediately when the patient exhibited shortness of breathing to ensure the airway is unobstructed. Mechanical ventilation was continued until the infection in the child's neck and chest was controlled and until the respiratory function recovered. After ensuring the unobstructed airway, the patient can accept various treatments. 2) The patient was given a combination of antibiotics when the diagnosis of DNM was considered. Although no evidence of bacterial cultivation was observed in the sensitivity test, effusion and gas appeared in the deep gap, and the use of broad-spectrum antibiotics exhibited a significant effect. 3) When the patient exhibited emphysema, symptomatic treatment of exhaustion and right pleural drainage were conducted at the same time. The CT scan after 1 month revealed that the left mediastinum had emphysema. The patient then underwent pleural drainage and left mediastinal incision drainage in two separated surgeries, which obtained significant results after 10 days. The patient's breathing improved, and the mediastinal and pleural drainage reduced and

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completely disappeared. 4) The chopstick-injury might have induced oesophagus rupture; hence, early nasogastric tube feeding was employed in the left mediastinal abscess. Analysis revealed that it might be caused by the oesophageal damage infection. The patient was fed milk with the nasogastric tube in long-term supine position. The milk might reflux from the stomach and aggravate the infection, leading to worse mediastinitis. Nasogastric feeding would easily cause reflux, residues and aspiration, whereas nasal jejunal feeding intubation would be deeper, causing diarrhoea, nutrient malabsorption and intussusception [25]. Nayak et al. [26] said that Nutritional supplements to the critically ill patients are one of the major issues to be discussed. Enteric feeding is advantageous over parenteral feeding because it maintains gut integrity and prevents bacterial translocation. Sajid et al. [27] point out that postpyloris (nasoduodenal and nasojejunal) (PP) feeding in intensive therapy unit patients reduces the gastric residual volume and risk of aspiration pneumonia. Postpyloris feeding is also superior to nasogastric feeding in terms of delivering higher proportion of daily caloric requirements. PP feeding with the help of nasoduodenal or nasojejunal tube may be used routinely in ITU patients for nutritional support. Doing so opens a valuable opportunity for the next step of treatment. In our case, the patient underwent second adjustment therapy using nasoduodenal feeding under the guidance of an endoscope. This strategy not only can avoid the reflux of the stomach contents to worsen the infections but also can provide the adequate enteral nutrition. At 10 days after duodenal feeding, the mediastinal infection was under control, the drainage of mediastinal effusion was significantly reduced and remarkable results were achieved. Therefore, gastroscopy-guided nasoduodenal feeding can prevent contaminants, such as oesophageal secretions and food, and enhance nutritional intake, providing better treatment for the rehabilitation of children with DNM.

Although the treatment lasted for more than 2 months, the result was satisfactory under active diagnosis and treatment of many physicians. The ill child was eventually discharged after restoring normal breathing and eating. The 3 month follow-up showed no abnormalities. Children with oropharyngeal trauma should receive complete treatment in a timely

manner. Although the wounds may not be obvious when the patient is admitted because of trauma, admitting doctors should be vigilant because the invisible closed wound may still cause serious deep cervical fascia infections and DNM.

Summary

The incidence rate of DNM caused by paediatric oropharyngeal trauma is low, but the mortality is high because the illness is dangerous and heavy. Thus, awareness should be raised for the suspected cases, and enhanced CT examination should be given in the early condition of patients to reduce missed and wrong diagnoses. After diagnosis, timely treatment should be performed, including the incision and drainage towards the chest-neck abscess, adequate application of broad-spectrum antibiotics and full symptomatic supportive treatment. For the suspected case of oesophageal perforation, fasting or nasoduodenal tube feeding should be administrated. The diagnosis and treatment of such cases involve the collaboration of otolaryngology, intensive care, radiology, thoracic surgery, internal medicine and other departments. All physicians should pay considerable attention to DNM to realise the early diagnosis and treatment, as well as improve the cure rate, of this disease.

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Disclosure of conflict of interest

None.

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