



Case Report

Multiple organ dysfunction due to a rare complication of Nuss procedure for pectus excavatum: A case report

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ARTICLE INFO

Article history:

Received 22 June 2020

Received in revised form

22 July 2021

Accepted 27 July 2021

Available online 5 August 2021

Keywords:

Multiple organ dysfunction syndrome

Nuss procedure

Acute cardiac tamponade

ABSTRACT

A 19-year-old male patient who suffered from sudden and repeated multiple organ dysfunction syndrome one month after the bar removal procedure of Nuss surgery for pectus excavatum was admitted to our department. With organ function supportive treatment, the etiology was finally identified to be a bone spur located at the inner border of the left costa due to repeated friction between the implanted steel bar and the rib, which damaged the heart repeatedly and induced the consequent acute cardiac tamponade. After operation, the patient was successfully managed and discharged. Follow-ups till three years indicated a good recovery.

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Introduction

Multiple organ dysfunction syndrome (MODS) or multiple organ failure is not uncommon in the intensive care unit (ICU), which can be secondary to internal disease or surgical intervention. The main and common inducing factors for MODS include sepsis and septic shock, severe acute pancreatitis, cardiopulmonary resuscitation, major surgery, and severe burns or trauma requiring surgical control. Patients with MODS have a high fatality. A 19-year-old patient with sudden MODS of unknown causes was successfully treated in our hospital. Our diagnosis and treatment for this patient once experienced food poisoning and infection, which troubled us for some time. Finally, it was found that the patient's symptoms were related to his Nuss surgical operation. We reported the diagnosis and treatment as follows.

Case report

A 19 years old male patient (Han nationality, student, unmarried) was admitted to our hospital on March 23, 2017 due to chest pain, oliguria for 10 days and phlegm for 1 week. He had a history of Nuss surgery 4 years ago (February 2013) due to pectus excavatum, and the metal bar for repair was removed in January 2017. The

patient had no other special medical history or family history. He felt nausea after drinking milk in the morning on March 13, with repeated violent vomiting of gastric contents, followed by chest pain.

The patient was sent to the local hospital immediately. Emergent electrocardiogram showed ST-segment elevation of the anterior partition. Coronary angiogram was performed immediately afterwards, which showed midsection of the anterior descending branch. Acute myocardial infarction and myocarditis was highly suspected and the patient was admitted to the cardiology department. On March 14, the chest pain symptoms were improved. But the patient still had nausea and vomiting and further progressed to anuria. Acute liver and renal failure was also considered, and thus the patient was transferred to the ICU that night. Emergency blood tests showed white blood cell (WBC) $18.3 \times 10^9/L$, neutrophils granulocyte (Grn)% 87.5%, glutamic-pyruvic transaminase 3469 U, glutamic oxalacetic transaminase 3815 U/L, total bilirubin 54.2 $\mu\text{mol/L}$, creatinine 217 $\mu\text{mol/L}$, urea nitrogen 10.8 mmol/L , serum potassium 7.47 mmol/L , creatine kinase 541 U/L, creatine kinase 50.4 U/L, lactate dehydrogenase 4138 U/L, procalcitonin (PCT) 0.13 ng/mL , C-reactive protein 22.8 mg/L , complement and protein 148. Anti-infection, continuous renal replacement treatment (CRRT) and other symptomatic treatments were given. Besides, blood, urine and other specimens were collected for etiological and toxicological examination. The patient's vital signs were gradually stabilized. On March 17, the patient had a sudden shock without obvious causes, followed by decreasing heart rate. Emergency endotracheal intubation, mechanical ventilation, cardiopulmonary resuscitation, and

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Peer review under responsibility of Chinese Medical Association.

fluid resuscitation were performed. As the blood pressure remained low, vasopressors were continuously given with 10 $\mu\text{g}/\text{kg}/\text{min}$ of Dopamine and 0.1 $\mu\text{g}/\text{kg}/\text{min}$ of Norepinephrine. After that, the vital signs returned stable again. Then the vasopressors were stopped on March 20 and the endotracheal intubation was removed on March 22.

Due to the difficulty in determining the pathogenesis, the patient was transferred to our hospital and admitted to the ICU on March 23. At the time of admission, he had a temperature of 36 °C, respiration rate of 24 breaths/min, pulse rate of 102 beats/min, and blood pressure of 154/98 mmHg. The patient had clear consciousness but in poor spirits. Skin and mucosa of the whole body were slightly yellow with scattered bleeding points, and subcutaneous ecchymosis could be observed in the limbs. The neck was soft with no resistance, the thoracic deformity was funnelform, and the old surgical scar can be seen on the chest. Other symptoms included increased respiratory mobility and respiratory frequency, coarse breath sounds in both lungs, moist rales and sputum sounds, neat heart rhythm, low-pitched heart sounds, weakened muscle strength and muscle tension of the limbs, and edema in both lower extremities. No other obvious abnormalities were observed. Laboratory examination showed WBC of $8.36 \times 10^9/\text{L}$, Grn% 95.4%, platelet 65 g/L, Troponin T 0.182 ng/mL, creatine kinase 7.590 ng/mL, myoglobin 835.3 ng/mL, and brain natriuretic peptide (BNP) 6265 pg/mL. Chest radiographs showed lung infection (Fig. 1A).

Admission diagnosis was considered to be: (1) pulmonary infection; (2) renal failure; (3) liver failure; (4) acute cardiac insufficiency, acute myocardial infarction, myocarditis and (5) poisoning?

At the beginning of admission, the patient's vital signs were relatively stable. He presented with abundant sputum and difficulty in sputum expectoration. CT scan (Fig. 1B) showed left pleural effusion and pericardial effusion. Echocardiography was routinely conducted.

At 16:30 on March 25, the patient suddenly complained of palpitation and dyspnea after defecating about 400 mL yellow mucous stools in bed. Sweating and cyanosis was present at the same time. Monitor showed the heart rate of 130–140 bpm, blood pressure 120–130/70–80 mmHg, and oxygen saturation (mask oxygen inhalation with 6 L/min) 85%–90%. Then the heart rate continued to rise. Intubation and mechanical ventilation were

conducted immediately, after which the oxygen saturation rose to 92%–94%, arterial blood gas oxygenation index of 86.6 mmHg and lactic acid of 4.3 mmol/L. Emergency bedside echocardiography examination showed obvious restricted ventricular diastole, stroke volume 17 mL, and diastolic pericardial fluid sonolucent area. The anechoic area of the post pericardium is 1.8 cm during end diastolic period (left ventricular long axis view), and the anechoic area of the left ventricular lateral pericardium is 1.0 cm (from the four chamber view) and 1.4 cm (from subcostal four chamber view). While it is 2.8 cm at the apical pericardium and 4.1 cm at the right ventricular lateral pericardium from subcostal four chamber view. Flocculent echo was found in the pericardial cavity, and no abnormal blood flow signal was detected in the pericardial cavity. At 21:00 thorax puncture and drainage was performed, with tawny turbid effusion discharged. At 23:39 pericardiocentesis was performed bedside. Due to thoracic deformity and difficulty in puncture, one drainage tube was indwelled, and later ultrasonography was performed to confirm that the drainage tube was located in the thorax, and about 200 mL dark red fluid was drained. After indwelling the chest catheter, the patient's symptoms were slightly relieved and the vital signs were more stable than before. At 9:00 on March 26, the patient was given pericardiocentesis guided by bedside ultrasonography again, and bloody fluid was continuously extracted (Fig. 2).

At the same time, coagulation function disorder with low prothrombin complex and low fibrinogen was found, so coagulation factors were added to correct the coagulation to normal. Then the patient maintained stable for one day. The fraction of inspired oxygen decreased from 100% to 50%. The toxicology and etiology tests sent by local hospital reported no abnormal results.

On March 27, after the patient turned over on bed, oxygen saturation suddenly declined again. Reexamination by echocardiography showed that there was still a large amount of effusion in the inferior wall of the right ventricle. Continued active bleeding could not be excluded. There was no obviously improvement after adjusting the position of pericardial drainage tube and supplementing coagulation factors.

Cardiologist, thoracic surgeon and radiologist were invited for consultation. Re-check of the chest CT (Fig. 3) showed that there was a sharp osteophyte at the inner edge of the left rib, which may be caused by long-term friction between the bar placed at Nuss surgery and the ribs. After removal of the bar, the osteophyte

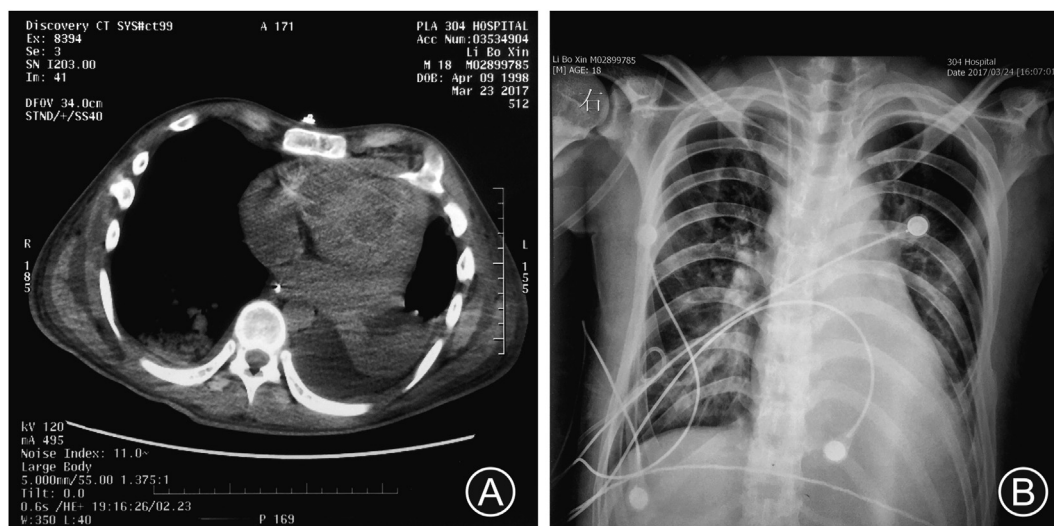


Fig. 1. (A) Chest radiograph on admission (March 24, 2017) revealed thickened lung texture; (B) Chest CT on March 23, 2017 showed left pleural effusion and pericardial effusion.

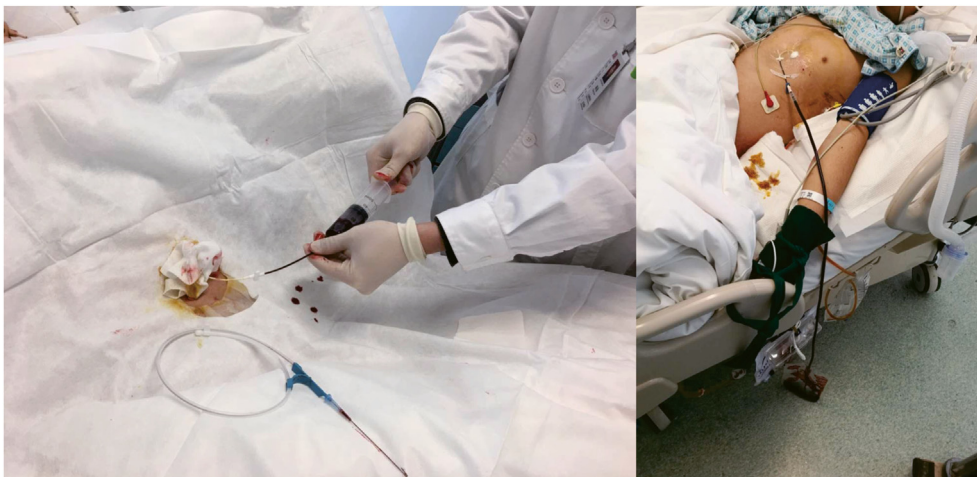


Fig. 2. Bloody pericardial effusion.

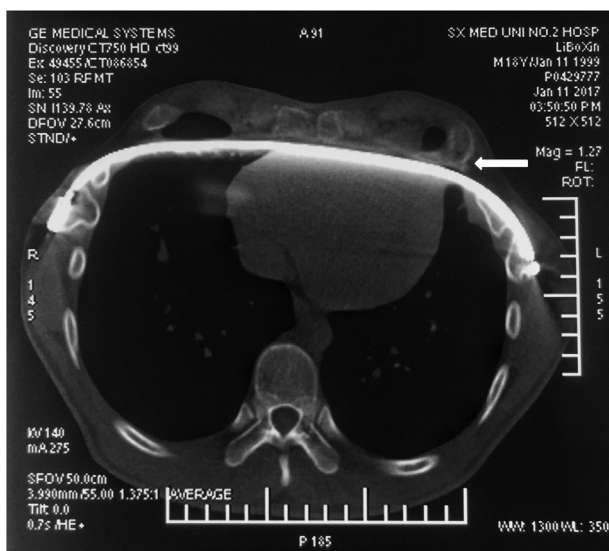


Fig. 3. Chest CT before plate removal on January 11, 2017. The arrow shows the sharp osteophyte.

directly and repeatedly damaged the pericardium, causing pericardial effusion and acute cardiac tamponage. Considering that the patient had repeated shock symptoms, affected functions of multiple organs, and complication of coagulation dysfunction, the surgical risk was considered to be extremely high. After repeated communication with his family, the patient underwent thoracotomy under general anesthesia at 18:00 on March 28.

During the operation, atelectasis of the lower left lobe with partial consolidation and about 500 mL of thoracic hydrothorax were revealed. The pericardium was partially adhered to the anterior chest wall; 300 mL blood fluid was extracted from the pericardium after pericardiotomy. Explored by pericardial window, an obvious damage was found to the anterior wall of the left ventricle, which was covered by fibrinoid material on the surface. Local blood exudation was observed after attempted cleaning, which was considered as the inducement of pericardial obstruction. The scar formation was about 3 cm long (Fig. 4A). Horizontal mattress suture was made using 4-0 suture with felt gasket. Osteophytes on the medial side of the ribs can be seen corresponding to the chest wall, with a sharp tactile sensation. The

osteophytes were trimmed and smoothed by a rongeur and wiped with bone wax. The costal osteophyte is shown in Fig. 4B.

The patient was returned to the ward after surgery. The drainage tube continued to draw bloody fluid. A large amount of fluid resuscitation and a small dose of vasopressor were needed to maintain the blood pressure, indicating active bleeding in the operative area. Urgent blood routine test showed hemoglobin was only 51 g/L, so another emergency surgical exploration was conducted 2 h after the operation. Diffuse extravasation was observed in the operative area. Extensive hemostasis was performed and coagulation factors were supplied. After the second operation, the patient was returned to the ward again and treated with mechanical ventilation, anti-infection and CRRT. Tracheotomy was performed on April 6, followed by intermittent blood supplementing, and the ventilator parameters was gradually turned down. Antibiotics were stopped on April 12. The renal function recovered, so the bedside CRRT was stopped on April 17, and rehabilitation exercise began. On May 16, the ventilator was taken off and the tracheal tube was removed. The patient was discharged on May 27. At the following follow-up after discharge, the patient was able to live and study normally, with no similar symptoms.

Discussion

MODS is very common in ICU wards. For MODS with definite primary disease, a better therapeutic effect is likely to be achieved by active treatment of the primary disease and providing organ function support. But for patients with unknown causes, the organ function may be repeatedly impaired even with a strong organ function support treatment. So the priority is to explore the cause to improve the prognosis.

Review of this case showed that the diagnosis and treatment protocols in the local hospital adhered to the conventional path, and the organ function support treatment and related screening were timely. After preliminary treatment, the condition was relatively stable. Based on the patient's specific "chest pain" symptoms and electrocardiogram changes as described in the medical history, though there was no ECG, coronary angiography was firstly performed in the local hospital. After that, acute myocardial infarction was excluded. The patient was an adolescent with a history of funnel chest but no other medical history. Considering that the patient drunk milk before the onset of severe gastrointestinal symptoms, together with the results of WBC and Grn% in blood routine, increased PCT and C-reactive protein, and the rapidly

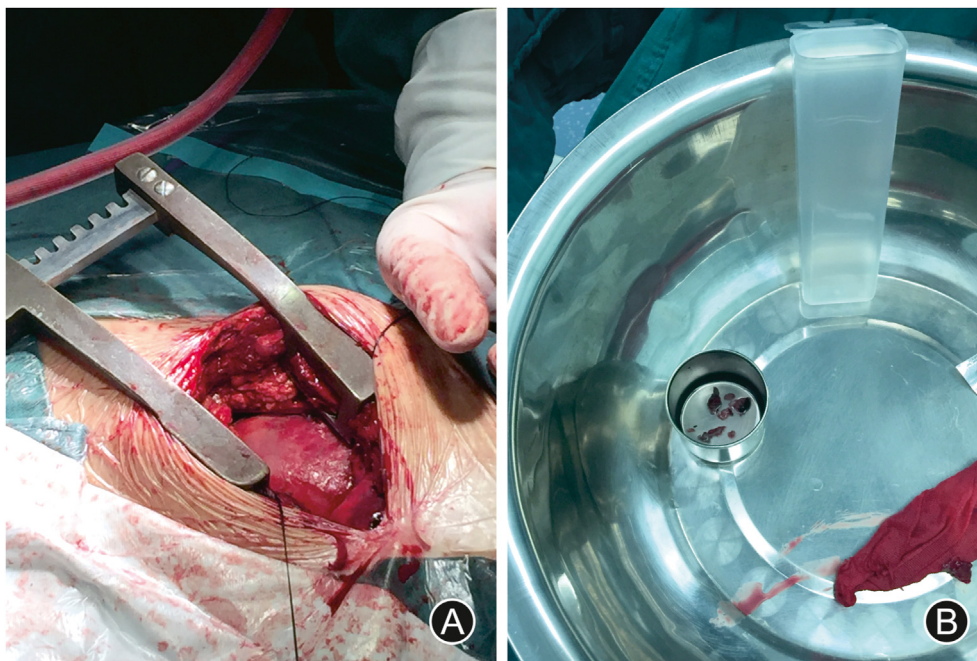


Fig. 4. (A) Scar on the pericardial surface; (B) Osteophytes proliferated in the left rib medial margin.

developed shock, liver failure and kidney failure on the same day, food poisoning from drinking tainted milk was first highly suspected in the local hospital. That was reasonable. But the subsequent negative toxicology tests precluded this diagnosis. After that, infection was considered in the local hospital, perhaps even in the sepsis stage, although the source of infection and etiology are unknown. The sequential organ failure assessment score was as high as 10 points, and the PCT increased, too. So the local hospital took sepsis as the priority suspected diagnosis.¹

Retrospectively, we can see that there are many reasons why the local hospital was unable to make a clear diagnosis. PCT is a sensitive indicator for infection. The patient's PCT was only slightly higher than the normal level while PCT below 0.25 g/L can exclude the possibility of bloodstream infection and abdominal infection.² Repeated exploration for the infection sites and repeated bacteriological tests did not reveal any presence of serious, life-threatening infections or explain the accompanying MODS. Mild elevation of PCT at the early stage may be associated with gastroenteritis. At this point, in the absence of a clear diagnosis of sepsis, other problems should be thoroughly screened. After the patient was admitted to the local hospital ICU, the sudden drop in blood pressure was clearly observed under the condition of ECG monitoring, which was relieved after resuscitation. Unfortunately, the local hospital had limited access to bedside echocardiography, so the real cause of circulatory disruption had not been found. Another restricted condition is that toxicology and etiological specimens need to be sent to Beijing for examination, and this time period is long, which would seriously affect the diagnosis and treatment process since the patient was critically ill.

After being transferred to our hospital, the relevant results were reported in turn to exclude poisoning, but the negative results of etiology could not completely exclude infection, so the most likely diagnosis in our hospital was also considered to be infection. The patient remained stable for some time, and the routine scheduled (non-urgent) echocardiography examination has not been completed within 2 days. When the patient suffered another

sudden change of condition, urgent echocardiography was performed and the diagnosis was thought to be acute cardiac tamponade. At that time, he has been complicated with hemodynamic instability and involved in respiration. Therefore, emergency pericardiocentesis was performed first to stabilize the vital signs temporarily.³ While the cause of acute cardiac tamponade was only conjecture, and subsequent surgical exploration confirmed this conjecture. After the surgical removal of the etiology, the patient's acute organ dysfunction was corrected successively and he finally returned to normal life.

Pectus excavatum is a common congenital thoracic deformity. Nuss is a minimally invasive infundibulum surgery for sternal malformation. Because of its advantages of minimally invasion, safety and aesthetics, Nuss has become the mainstream surgical method for the treatment of pectus excavatum. Life-threatening complications of Nuss surgery include perforation of the heart and injury of the great vessels. Although heart injury is extremely rare, it can endanger the life of the patient once occurs, with many existing reports.^{4–7} The serious complications of Nuss surgery are mostly occurred during the operation of bar insertion and removal, and postoperative complications are rare. Heart injuries related to Nuss are mainly caused by intraoperative expansion forceps when passing through the posterior sternal blind area, postoperative bar displacement, or during bar removal surgery. Intraoperative expansion forceps damaging the heart is the most common.⁸ Our patient underwent successful bar removal. Due to the small range of rib activity after the operation, there was no fatal injury to the pericardium, and the patient maintained a normal life for more than 2 months. An accidental cause (severe vomiting after drinking spoiled milk) led to a major change in thoracic morphology, with the sharp osteophyte piercing into the pericardium, followed by pericardial bleeding and tamponading. The injury may only affect the myocardium, but not the blood vessels of the heart, so the clinical manifestation was symptomatic pericardial tamponade after the change of thoracic position. The complication occurred

nearly 2 months after bar removal. This is extremely rare and no similar cases have been reported.

The original diseases of patients admitted to the comprehensive ICU ward were widely distributed. On the basis of organ function support, intensivists should actively seek for the cause and provide treatment accordingly to improve the final prognosis. In this process, multidisciplinary cooperation is very important. The diverse accessory examination equipment and physician experience and ability of each hospital also affect the therapeutic effect.

Funding

Nil.

Ethical statement

Informed consent has been obtained from the patient.

Declaration of competing interest

The authors declare no conflicts of interest or financial interests.

References

1. Seymour CW, Liu VX, Iwashyna TJ, et al. Assessment of clinical criteria for sepsis: for the third international consensus definitions for sepsis and septic shock (sepsis-3). *J Am Med Assoc.* 2016;315:762–774. <https://doi.org/10.1001/jama.2016.0287>.
2. Suberviola B, Castellanos-Ortega A, González-Castro A, et al. Prognostic value of procalcitonin, C-reactive protein and leukocytes in septic shock. *Med Intensiva.* 2012;36:177–184. <https://doi.org/10.1016/j.medin.2011.09.008>.
3. Ristić AD, Imazio M, Adler Y, et al. Triage strategy for urgent management of cardiac tamponade: a position statement of the European society of cardiology working group on myocardial and pericardial diseases. *Eur Heart J.* 2014;35:2279–2284. <https://doi.org/10.1093/eurheartj/ehu217>.
4. Moss RL, Albanese CT, Reynolds M. Major complications after minimally invasive repair of pectus excavatum: case reports. *J Pediatr Surg.* 2001;36:155–158. <https://doi.org/10.1053/jpsu.2001.20039>.
5. Gips H, Zaitsev K, Hiss J. Cardiac perforation by a pectus bar after surgical correction of pectus excavatum: case report and review of the literature. *Pediatr Surg Int.* 2008;24:617–620. <https://doi.org/10.1007/s00383-007-2097-1>.
6. Becmeur F, Ferreira CG, Haecker FM, et al. Pectus excavatum repair according to Nuss: is it safe to place a retrosternal bar by a transpleural approach, under thoracoscopic vision. *J Laparoendosc Adv Surg Tech.* 2001;21:757–761. <https://doi.org/10.1089/lap.2011.0035>.
7. Scharschmidt K, Lempe M, Schlesinger F, et al. Lessons learned from lethal cardiac injury by Nuss repair of pectus excavatum in a 16-year-old. *Ann Thorac Surg.* 2013;95:1793–1795. <https://doi.org/10.1016/j.athoracsur.2012.09.038>.
8. Bouchard S, Hong AR, Gilchrist BF, et al. Catastrophic cardiac injuries encountered during the minimally invasive repair of pectus excavatum. *Semin Pediatr Surg.* 2009;18:66–72. <https://doi.org/10.1053/j.sempedsurg.2009.02.002>.