CASE REPORT

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Spontaneous air leak syndrome – a very rare pulmonary complication of an eating disorder

Sindrom spontanog curenja vazduha – veoma retka plućna komplikacija poremećaja u ishrani

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Abstract

Introduction. Eating disorders are characterized by persistent body dissatisfaction and unhealthy weight control. Spontaneous air leak syndrome (ALS) is a rare but potentially fatal complication of a perennial eating disorder that can lead to malnutrition with hypoproteinemia. We present a young male suffering from anorexia nervosa who developed a severe form of spontaneous ALS. **Case report.** The 25-year-old male patient was initially treated for problems with an eating disorder of the persistent anorexia nervosa type. He deliberately lost about 40 kg during the previous four years. During hospital treatment, the patient suddenly developed severe pain in the abdomen, for which he was urgently referred to a surgeon. He was admitted to the Intensive Care Unit exhausted and afebrile, with the abdomen below the level of the chest and diffuse painful tenderness, with peristalsis audible and petechiae present on the skin of the back, arms, and thoracic wall. Computed tomography of the thoracoabdominal region was performed, which showed air in the mediastinum, in the spinal canal, around the stomach, in the rectum, and in the small pelvis. After an emergency laparotomy, a drainage of the abdominal cavity and intraoperative esophagogastroduodenoscopy were performed. The patient developed neurological complications postoperatively. After a successful recovery, he was treated psychiatrically on an outpatient basis and monitored for several months. Further similar complaints did not occur. **Conclusion.** Patients with nutritional disorders should be closely monitored because a significant protein deficit with spontaneous ALS can occur. Timely diagnosis and treatment can prevent further somatic deterioration and save the patient's life.

Key words:
anorexia nervosa; diagnosis; emphysema; feeding and eating disorders; laparotomy; surgical procedures, operative; tomography, x-ray computed.

Apstrakt

Uvod. Poremećaje u ishrani odlikuju uporno nezadovoljstvo sopstvenim telom i nezdrava kontrola telesne mase. Sindrom curenja vazduha (SCV) koji se javlja spontano je retka ali potencijalno fatalna komplikacija višegodišnjeg poremećaja u ishrani, koji dovodi do pothranjenosti sa hipoproteinemijom. Prikazujemo mladića lečenog od anoreksije nervoze, kod koga se razvio težak oblik spontanog SCV. **Prikaz bolesnika.** Bolesnik star 25 godina je prvobitno lečen zbog problema sa poremećajem u ishrani po tipu uporne anoreksije nervoze. Namerno je izgubio oko 40 kg tokom prethodne četiri godine. Tokom bolničkog lečenja, bolesnik je iznenada dobio jak bol u trbuhu, zbog čega je hitno upućen hirurgu. Na odeljenje intenzivne nege primljen je iscrpljen, afebrilan, sa trbuhom ispod nivoa grudnog koša, difuzno bolno osetljivim, sa čujnom peristaltikom i petehijama na koži leđa, ruku i zidu grudnog koša. Urađena je kompjuterizovana tomografija torakoabdominalne regije, kojom je viđen vazduh u medijastinumu, u kičmenom kanalu, oko želuca, rektuma i u maloj karlici. Posle hitne laparotomije, urađena mu je drenaža trbušne duplje i intraoperativno ezofagogastroduodenoskopija. Bolesnik je postoperativno imao neurološke komplikacije. Posle uspešnog oporavka, ambulantno je psihijatrijski lečen i praćen tokom više meseci. Nadalje se nisu javljale slične tegobe. **Zaključak.** Bolesnike sa poremećajima u ishrani treba pažljivo pratiti, jer kod njih može doći do značajnog deficita proteina, sa spontanim SCV. Blagovremena dijagnoza i lečenje mogu sprečiti dalje somatsko pogoršanje i spasiti život bolesnika.

Ključne reči:
anoreksija nervoza; dijagnoza; emfizem; ishrana, poremećaji; laparotomija; hirurgija, operativne procedure; tomografija, kompjuterizovana, rendgenska.

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Introduction

Eating disorders are characterized by a persistently disturbed eating pattern and perception of body appearance, which results in significant impairment of psychological health and social functioning. A disturbed eating pattern includes skipping meals, restriction of energy intake, or intake of specific macronutrients, with or without vomiting, further complicating the disease's clinical course [1]. Emphysema is a pathological loss of alveolar tissue with increased air space and consequent respiratory function disorder. When it occurs in patients with eating disorders, it presents a serious complication that requires immediate care. In the 1970s, emphysema research was started on animal models of anorexia. The results of initial research indicate that in starved rats, there is a reduction of surfactant with an increase in intraalveolar tension and a tendency towards lung collapse. Surfactant is surface-active fluid/liquid rich in proteins and lipids, and it is produced by alveolar cells to maintain adequate surface tension in the alveoli and the resulting gas exchange at the level of the alveocapillary membrane. Starved rats had reduced protein synthesis and increased proteolysis [2, 3].

Clinical studies have shown that in patients suffering from eating disorders, there is an increase in proteolysis, which, together with previously used lipids, become the material for energy metabolism. Due to the general energy deficit and hypoproteinemia, there is weakness in the respiratory muscles and the functionality of the diaphragm, with systemic oxygen hypoperfusion [4]. Spontaneous air leak syndrome (ALS), characterized by the development of emphysema with pneumothorax, pneumomediastinum, and/or pneumoperitoneum, is a rare but life-threatening complication. It is associated with eating disorders, primarily anorexia nervosa (AN). The syndrome is a consequence of prolonged malnutrition with hypoproteinemia, alveolar damage, and subsequent emphysema leading to additional complications [5]. In this regard, every patient suffering from AN who has respiratory problems or sudden unexplained pain in the abdomen should be taken seriously and examined somatically. Patients who often vomit and who otherwise have a history of frequent respiratory diseases or chronic diseases such as asthma and bronchitis are at particular risk. Pneumomediastinum, subcutaneous emphysema, epidural emphysema, and interstitial emphysema have also been reported in severe AN cases [6, 7].

Although the medical complications of AN are numerous, pulmonary complications are still very rare. Although the exact pathogenetic mechanism has yet to be determined, we can agree that these complications in our patient are secondary and caused primarily by permanent nutritional starvation.

Case report

In our study, we present a 25-year-old male patient, who initially came for an examination in May 2019 due to symptoms of significant body mass loss (he lost 32 kg) in a period of three years (from 2015 to 2018). Due to the need for additional examination, he was admitted to the Department of Adolescent Psychiatry and Psychotherapy at the Clinical Hospital Center "Dr. Dragiša Mišović-Dedinje", Belgrade, Serbia.

The patient is the second of two children from a complete family. The mother's pregnancy and delivery went well, as did the patient's early psychomotor development. He went to school on time and achieved very good and excellent results.

Problems in terms of eating disorders started early in life. During most of his life, the patient was obese, but he had no problems in social relations. After a long, stressful life period, the patient weighed 120 kg in 2015. Then, he started to feel tired in his daily activities, so he decided to lose weight. He started exercising intensively, changed his diet in terms of eating healthy food and regular meals, and reduced his total daily caloric intake. From December 2018 until he reported to the psychiatrist (five months later), he lost 20 kg. In December, while preparing for the last exam, he lost 3–4 kg and regained weight during the New Year holidays. From 2015 to 2018, he lost a total of 32 kg, which, considering his height, was a lot, and at the time of admission, it was life-threatening.

At the time of admission to the Department of Adolescent Psychiatry and Psychotherapy in 2019, he was visibly cachectic, preoccupied with thoughts about food, physical appearance, and his own health. In the first act of treatment, the patient was in the hospital for three days. He left at his own request. The second admission to the Department was realized one month after the previous discharge, in June 2019, after the patient lost another 10 kg (body weight 53 kg, height 187 cm, body mass index 15.156 kg/m^2). On the very day of admission, in order to "prove" that he will start eating adequately and that he does not have to be hospitalized, he ate a large amount of pastries and other high-calorie foods. On the day of admission, in the afternoon, the patient began to complain of severe, diffuse pain in the abdomen that did not stop. Because of the above, a consultation with a surgeon and further somatic treatment was performed.

After a general surgeon examination, the patient was admitted to the Intensive Care Unit. Somatic status showed that the patient was exhausted and afebrile, with the abdomen below the level of the chest and diffuse painful tenderness, peristalsis audible and clear and petechiae present on the skin of the back, arms, and thoracic wall. A computed tomography (CT) scan of the abdomen and pelvis was indicated. The CT scan showed air in the mediastinum and spinal canal. In the arterial phase, there was a filling defect of the *truncus brachiocephalicus*, which could correspond to a thrombus. Pronounced collaterals behind the spinal column were also noted, and air was present around the stomach, rectum, and in the small pelvis (Figures 1–4).

The patient was treated operatively – an exploration of the abdominal cavity with drainage and intraoperative esophagogastroduodenoscopy was performed. Preoperatively,

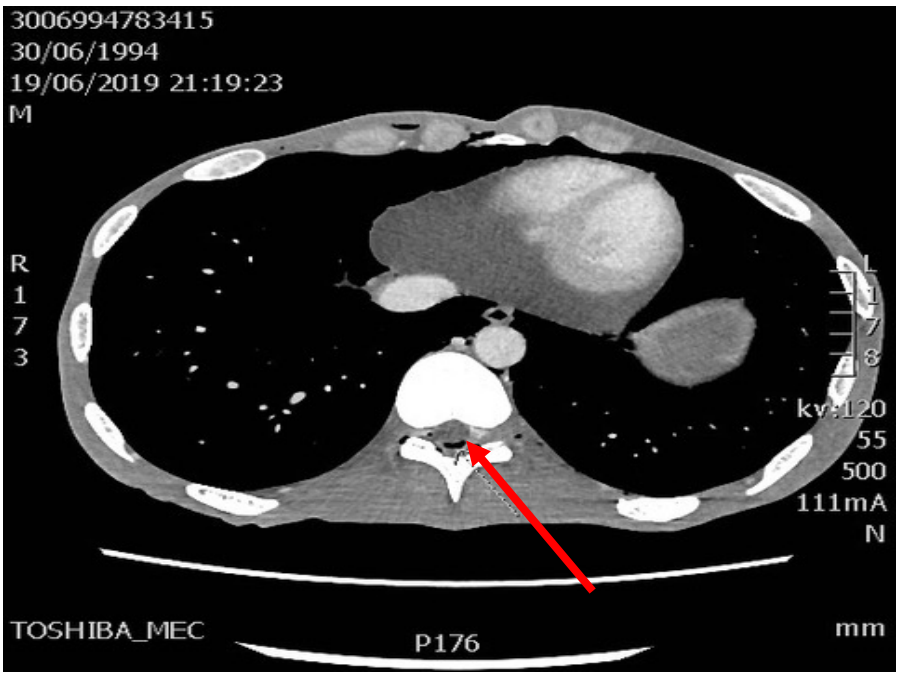

Fig. 1 – Computed tomography scan shows air in the spinal canal (red arrow indicates change).

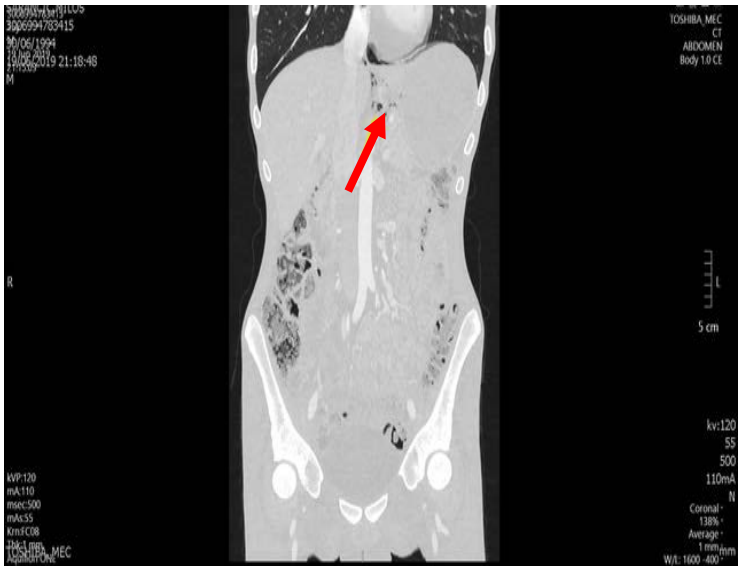

Fig. 2 – Computed tomography scan shows air retroperitoneally (red arrow indicates change).

minimal duodenal hemorrhage and cardiac insufficiency were noted. Postoperatively, the patient had a prolonged recovery with additional complications. Physical therapy, breathing exercises, and gradual verticalization were prescribed. Due to the catheterization and general poor physical condition, he developed a urinary infection and a bilateral hydrocele. A neurologist was consulted due to a foot drop. On the clinical examination, the following was found: a reduced trophism on the upper extremities, decreased gross motor power, reduced trophism on the lower extremities, no ability to dorsiflex the foot to the left, decreased patellar and Achilles reflexes, no ability to perform Mingazzini's test, Lazarevic's sign positive bilaterally at 110 degrees, left fanned plantar response. Investigation for porphyria was performed but returned negative. Magnetic resonance imaging of the lumbosacral part of the spinal column was performed, and the findings showed the existence of a focal change in the projection of the left transverse process of the second lumbar (L2) spinal vertebral body, which would be a differential diagnosis in favor of a hemangioma. During hospital treatment, laboratory analyses were performed on several occasions, in which elevated parameters of inflammation and hypoproteinemia dominated. After rehabilitation, one month later, the patient was discharged in a better general condition. A selective serotonin reuptake inhibitor (paroxetine) in a dose of 20 mg in the morning and olanzapine were prescribed as psychopharmaceuticals. Follow-up was continued for another year, after which the patient had no new somatic

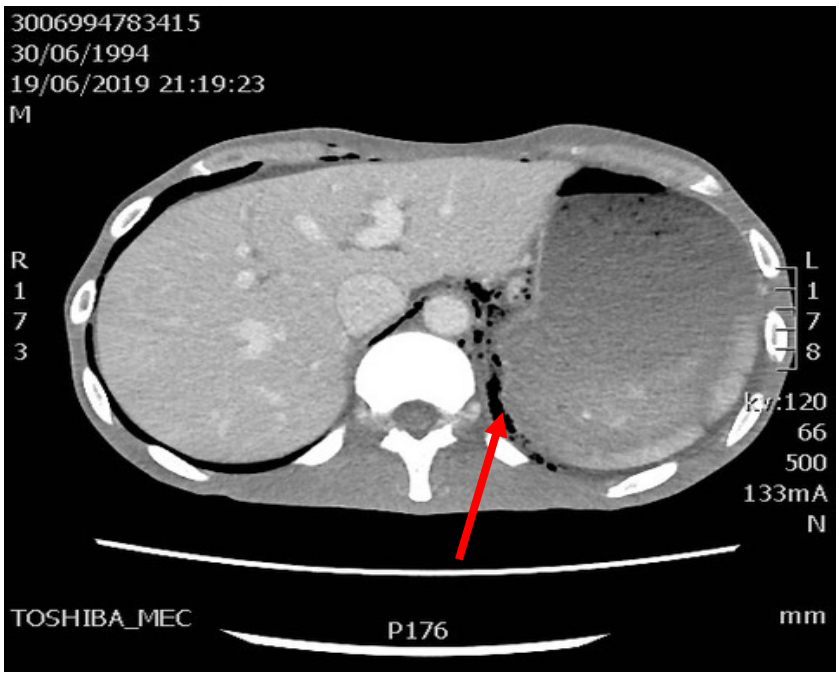

Fig. 3 – Computed tomography scan shows pneumoretroperitoneum (red arrow indicates change).

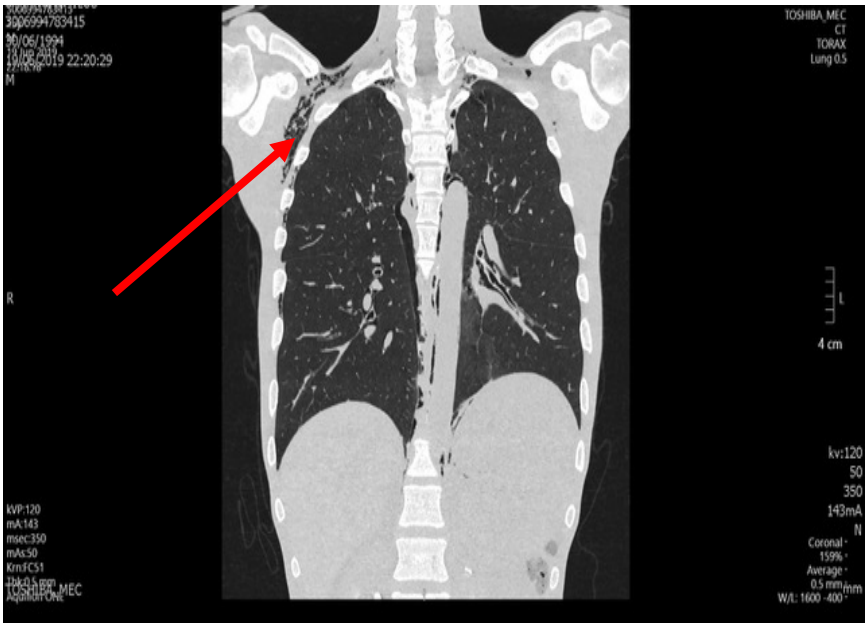

Fig. 4 – Computed tomography scan shows pneumomediastinum (red arrow indicates change).

complications. Since an adequate nutritional intake was established, there was a restoration of body mass as well as an improvement of psychological functioning. After thi, the patient did not come for follow-up psychiatric examinations.

Discussion

Eating disorders, predominantly AN, carry numerous somatic complications. Although pulmonary complications are rarely described, we believe that in our patient, ALS, with the consequent development of pneumothorax, pneumomediastinum, and pneumoperitoneum, arose as a result of permanent long-term nutritional restriction with a significant protein deficit.

Insufficient protein intake with hypoproteinemia leads the organism to a state of proteolysis. This further leads to the reduction of surfactant, an alveolar fluid that is rich in proteins and lipids and has the function of maintaining adequate surface tension in the alveoli. With the reduction of surfactant, alveolar collapse occurs, which in severe cases further leads to rupture of the alveolar walls with the development of emphysema. This emphysema was named “nutritional emphysema” since the main cause in its genesis is malnutrition [8, 9]. The condition can be complicated further

with the development of pneumothorax and pneumomediastinum. From the mediastinum, air leaks further to the peritoneum *via* the fascial planes of the perivascular space. It is known that the neck and mediastinum communicate with the peritoneum and retroperitoneum through a common visceral space that surrounds the esophagus and trachea and then follows the esophagus through the diaphragmatic opening. There is also an explanation that the presence of a diaphragmatic hernia, combined with anatomical variations where the parietal layer of the peritoneum is missing, leads to the communication of the peritoneum with the mediastinum. These sites of potential communication are called the *foramina* Morgagni, a parasternal defect of the diaphragm, and the *foramina* Bochdalek, a posterolateral defect of the diaphragm [10, 11].

The observed research findings in experimental animal models have been confirmed clinically to some extent. In order to demonstrate the mechanism of elasticity reduction, the content of connective tissue was examined in starved and normally fed rats. In starved animals, a significantly lower amount of connective tissue, hydroxyproline, elastin, and protein was found in the lungs. With the resumption of regular feeding, the hydroxyproline content normalized, but the protein, connective tissue, and elastin content were still at a level lower than normally expected. This showed that, in addition to the reduction of surfactant, changes in the lung tissue can also be explained by the loss of connective tissue [12]. The observed findings in experimental animals have been clinically confirmed to some extent [5].

The risk of developing spontaneous pneumothorax, pneumoperitoneum, and pneumomediastinum is present in all patients with persistent restrictive food intake, especially in patients who vomit due to increased intrathoracic and intraabdominal pressure, which triggers a cascade reaction at the level of previously present alveolar damage [13].

The presented patient had an extremely restrictive caloric intake for a long time. For some time, before being admitted to the hospital, he tried to gain weight by consuming a large amount of caloric food. Because of that, a sudden onset of stomach pain occurred, causing suspicion of somatic complications from the gastroenterological domain. However, after performing imaging diagnostics and obtaining information that the patient has a pneumothorax with pneumoperitoneum and pneumomediastinum, the diagnosis was made in the direction of ALS, a rare but potentially fatal complication in patients who have been exposed to a nutritional deficit for a long time, especially in eating disorders characterized by a long term pattern of starvation and malnutrition.

A phenomenon called the McLean effect has been described in which air trails/leaks after alveolar rupture spread along the bronchovascular bundle back to the mediastinum, leading to pneumomediastinum [13]. Pneumothorax, as a spontaneous secondary phenomenon, has also been reported in avoidant/restrictive food intake disorder - ARFID. This has also been linked to a nutritional deficit, although in this type of eating disorder, body weight may be within the normal range or even excessive. Furthermore, a higher frequency of chronic pulmonary diseases, such as asthma and cystic fibrosis, has been shown in cases of bowel disorders. Namely, these chronic, debilitating diseases were more often associated with inadequate nutrition, lower caloric intake, malnutrition, and, in predisposed cases, with the development of eating disorders [14, 15].

ALS has been described as a complication of numerous internal diseases [bleomycin-induced pneumonia, coronavirus disease 2019 (COVID-19) pneumonia, bronchiolitis obliterans, systemic sepsis, rheumatoid arthritis] and conditions (foreign body in the bronchus, traumatic nasolaryngeal suction), in premature babies with acute respiratory distress syndrome, after transplantation as part of the graft *versus* host disease reaction, and in patients with human immunodeficiency virus [16–20]. However, as a complication of psychiatric disorders, ALS has been described in a very small number of patients with AN. Our case presentation is in positive correlation with the results of other clinical studies stating that vomiting is a rare cause of the development of ALS with consequent pneumothorax. In other research, as well as in our report, it was shown that patients with AN recover more slowly from this complication compared to patients with other diseases with the same complication. Moreover, some papers write about the recurrence potential of ALS in patients with AN due to lung tissue damage, which was not the case in our patient [21, 22].

Conclusion

Eating disorders are among the leading causes of mortality among psychiatric phenomenology. However, the fatal outcome is primarily a consequence of permanent starvation or the act of suicide in these patients. Spontaneous air leakage syndrome, as a potentially fatal complication of permanent malnutrition, should be taken seriously, diagnosed urgently, and promptly treated.

The importance of the presentation of this clinical case is primarily reflected in spreading awareness that eating disorders, although they have a core psychiatric component, are systemic disorders with potentially disabling and fatal complications, which makes them one of the most serious disorders in psychiatry and medicine in general. The importance is also reflected in the fact that such complications are very often not diagnosed in clinical practice because sometimes the symptoms do not have to be dramatic, and the consequences can remain and lead to other somatic problems later in the patient's life. It is always necessary to think about pulmonary complications in any patient suffering from an eating disorder who experiences pain in the region of the neck and chest, difficulty breathing, dysphagia, and abdominal pain. Furthermore, even in cases where the listed symptoms are absent, one should think about this complication of the underlying disease because timely, adequate diagnosis and therapy can prevent further somatic damage and save the patient's life.

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