PERSISTENT PNEUMOTHORAX IN A PREMATURE INFANT

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ABSTRACT

Introduction: Pneumothorax is a condition in which the air is present in the pleural space with a collapse of the ipsilateral lung. Spontaneous pneumothorax is divided into primary and secondary. Primary pneumothorax makes 80% of all pneumothoraxes and there are no pathological changes present in the lungs. Secondary pneumothorax is a condition in which there is a lung disease in the background. A rare cause of secondary pneumothorax in premature infants is a bronchopleural fistula, which is a pathological communication between the bronchi and the pleural space.

Objective: Indicate the need for early diagnosis and evaluation of secondary pneumothorax in order to carry out the early optimal treatment of the patient.

Case report: Male premature infant, born from a high-risk pregnancy, gestational age of 31+6/7 weeks, low birth weight, presented with respiratory failure due to the respiratory distress syndrome, and persistent pneumothorax with the accompanying ventilation difficulties. Given the relapsing nature of the disease and limited compensatory mechanisms in premature neonates, a thoracotomy was indicated and a bronchopleural fistula of the upper section of the right lung was verified. The congenital pathological communication between the bronchi and pleural space was corrected by surgical procedure. The procedure went well same as the postoperative recovery on the department.

Conclusion: Congenital bronchopleural fistulas are rare and big issue in premature neonates. Clinical sign, suggesting a bronchopleural fistula, is a persistent and relapsing pneumothorax, which - if left untreated - would lead to the obstructive shock and heart failure due to the collapse of the compensatory mechanisms. Rapid diagnostic and therapeutic approach is important for having a good outcome for our patients.

Key words: persistent pneumothorax, bronchopleural fistula, secondary pneumothorax, premature infant.

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INTRODUCTION

Spontaneous pneumothorax is divided into primary and secondary. Primary pneumothorax makes 80% of all pneumothoraxes and there are no pathological changes present here in the lungs. Secondary pneumothorax is a condition in which the etiologic causes of pneumothorax (1, 2) may be established by X-ray methods. At the neonatal age, the secondary pneumothorax most often appears as the complication of the respiratory distress syndrome, aspiration of meconium or congenital malformations, which is deteriorated by mechanical ventilation (3). Persistent pneumothorax is a rare and demanding condition in neonatology, often followed by high mortality and morbidity rate. Permanent air leak to thoracic drain suggests the existence of bronchopleural fistula. Bronchopleural fistula is a pathological connection, assuring the communication between bronchi and pleural space with permanent air leak for more than 48 hours after having the thoracic drainage placed (4). Since the persistent pneumothorax is a rare clinical event in the neonatal age, the uniform therapeutic guidelines have not been agreed and defined yet in the management and evaluation of the premature infants with bronchopleural fistula.

CASE REPORT

Male premature infant, gestational age of 31+6/7 weeks, low birth weight of 1890 grams, admitted at the Department of Intensive Paediatric Treatment immediately after the urgent birth-delivery by Caesarean section due to the hypertensive crisis of a mother. The child was born from the third pregnancy, complicated by hypertension, pneumonia, machine ventilation and Langerhans histiocytosis of the mother. At admission, the child was breathing independently with moderate respiratory efforts and initial signs of the respiratory insufficiency. Oxygen therapy was applied via nasal prongs, but the respiratory insufficiency prograded with respiratory acidosis and hypercapnia. The X-ray images verified the changes in the lungs in terms of the respiratory distress syndrome. A surfactant was applied by endotracheal airway and a non-invasive machine ventilation was started by biphasic positive airway pressure in lungs (BiPAP). A mixed acidosis persisted in the acid-base status of blood. Dobutamine was introduced in the therapy with empirical antibiotics prophylaxis (ampicillin, amikacin, ceftazidime). The patient was given vitamin K and the parenteral feeding was started with small volumes of enteral nutrition. Ultrasound examination of brain detected the intracranial haemorrhage of the 3rd degree, which additionally aggravated the already complicated hospital course. On the second day of the child's life, the respiratory function deteriorated, the patient was analgosedated and intubated and the child started breathing via invasive machine. The X-ray diagnostics confirmed the displaced shadow of cardio mediastinum on the left side and suspected pneumomediastinum along with the previously described changes in the lung parenchyma. Laboratory tests recorded the increase of inflammatory parameters and signs of the early perinatal infection. Antibiotic therapy

was corrected based on the received microbiological test results and meropenem was included. The following days of the child's stay were characterised by difficulties with ventilation and verification of the right-side of the pneumothorax. The paediatric surgeons were consulted and the intrathoracic drain was placed in the right pleural space. Despite the afore-mentioned, the drainage of the pneumothorax was insufficient and aggravated and the keeping of oxygenation was aggravated as well, which led to the multiple drainage of the right chest, with no success unfortunately. It was agreed to connect the patient to the thoracic pump with negative pressure, whereupon a short-term apparent improvement occurred in clinical features. By obstruction of the drain, few days later the discomfort difficulties repeated. Computed tomography of lungs (MSCT) confirmed the existence of the pneumothorax on the right side with cystic expansion of bronchi on peripheral side in the medium and lower field of the lung. With regard to the relapsing pneumothorax and the impossibility of achieving the re-expansion of lungs, the thoracotomy was indicated in the surgery facilities of the Paediatric Surgery Clinic of the University Clinical Hospital Mostar (UCH).

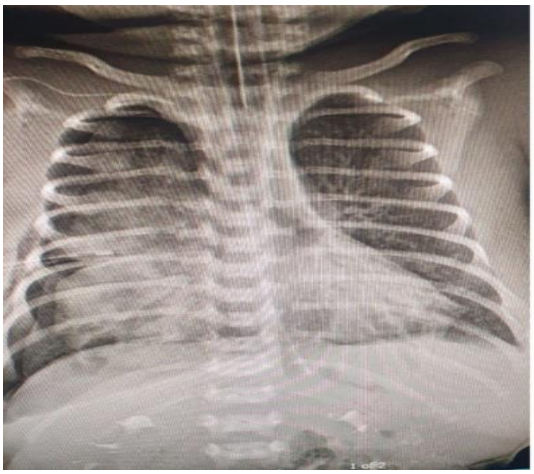

Image 1. The X-ray image shows the respiratory distress syndrome (RDS) and pneumothorax in the 2nd day of life with visible shadows of tube, intrathoracic drain and central venous catheter on the right side.

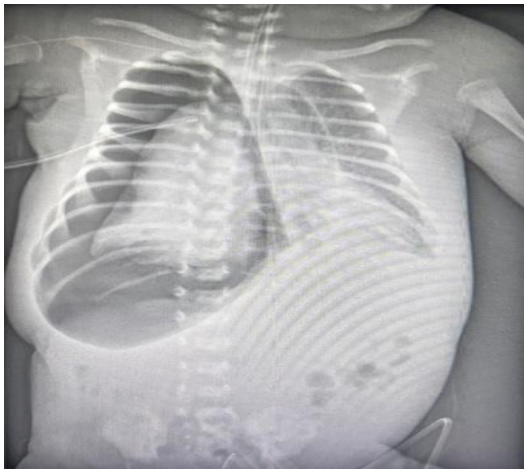

Image 2. The X-ray image shows the persistent right pneumothorax during the stay.

A surgery was carried out under general endotracheal anaesthesia. During the surgery, the decortication was carried out and major bronchopleural fistula sutured at the upper right section of the lungs. The surgery went well. An intrathoracic drain was placed and the wound was stitched up per layers. The patient was returned to

the Department of Intensive Paediatric Treatment after surgery, where a favourable post-surgical course took place with gradual separation from the machine ventilation and removing the intrathoracic drain out on the fifth day post-surgery. Further recovery went normally, the patient had a normal clinical and laboratory status and was achieving a good progress in weight. The child was discharged from hospital for home care after two months of hospital treatment.

DISCUSSION

A persisting pneumothorax represents a great challenge in therapeutic approach and according to the literature, several therapy solutions have been tried so far. The treatment with insertion of thoracic drain was most often carried out, but it was also tried with selective bronchial occlusion, pleurodesis with talc, tetracyclines and fibrin glue (4, 5). For newborn children a flexible drain of 6 - 8 French was used for drainage, and in about 25 % of premature infants the complications were recorded during the drainage in the form of the obstruction of catheter, rupture of drains, extraction and failed drainage (6). In case of the persistent pneumothorax, the clinical features of respiratory distress were developed, and in acid-base status of blood the hypoxia was always with the test result of hypercapnia, which finally led to the respiratory insufficiency and severe global hypoxia (7, 8). All modes of treatment had the same purpose and it is the re-expansion of lungs and setting up of normal ventilation. In cases, when in addition to active suction, the pneumothorax would persist for more than 48 hours, some authors had good experience with pleurodesis. The premature infants with small birth weight are a special challenge in treatment because in this population urgent surgery was often indicated in case of a bad response to multi-drainage insertions. Our patient was one of such cases. Although such cases of treatment are described, the routine use of fibrin glue is not recommended in the treatment of bronchopleural fistula because of the side-effects, because it causes bradycardia, hypercalcemia, diaphragmatic paralysis, and counter-lateral pneumothorax. Fibrin glue was indicated only in case of absence at conventional treatment (5). Chemical pleurodesis seems to be more likely the option for treatment of this condition in adults. Our patient was treated by insertion of intrathoracic drains and light suction of pneumothorax via thoracic pump (10-15 cm H_2O). Because of the bad response to that therapy attempt and repeated development of pneumothorax, the thoracotomy was carried out as well as the saturation of bronchopleural fistula, after which the clinical condition significantly improved. Since it is about the clinical rarity at the neonatal age, especially at the premature infants, a few isolated clinical experiences are available in the literature, and the therapy solutions were mainly reduced down to the insertions of multiple thoracic drains along with a slow suction of air via thoracic pump (10-15 cm H_2O) (4).

CONCLUSION

Pneumothorax is a known entity in the neonatal age, especially in patients with machine-induced breathing. However, the congenital bronchopleural fistulae are rare and represent a great problem at this age. A clinical sign, suggesting the bronchopleural fistula, is a persistent and relapsing pneumothorax, which if not treated - leads to the obstruction shock and heart failure due to the collapse of the compensatory mechanisms. Timely determination of an accurate diagnosis and fast surgical intervention are extremely important in the treatment of these congenital malformations of the respiratory system. Thanks to the good surgical – paediatric approach, our patient had a favourable treatment outcome.

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PERZISTENTNI PNEUMOTORAKS KOD NEDONOŠČETA

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SAŽETAK

Uvod: Pneumotoraks je stanje u kojem je prisutan zrak u pleuralnom prostoru s kolapsom ipsilateralnog plućnog krila. Spontani pneumotoraks se dijeli na primarni i sekundarni. U primarni spada 80% svih pneumotoraksa i kod njih nema patoloških promjena u plućima. Sekundarni pneumotoraks je stanje u kojem se radiološkim metodama mogu utvrditi etiološki uzroci nastanka pneumotoraksa, dakle pneumotoraks u podlozi ima plućnu bolest. Rijedak uzrok sekundarnog pneumotoraksa u nedonoščadi je bronhopleuralna fistula koja označava patološku komunikaciju između bronha i pleuralnog prostora.

Cilj rada: Ukazati na potrebu za ranom dijagnozom i evaluacijom sekundarnog pneumotoraksa kako bi se što ranije proveo optimalan terapijski tretman pacijenta.

Prikaz slučaja: Radi se o muškom nedonoščetu, rođenom iz komplicirane trudnoće, gestacijske dobi 31+6/7 tjedana, niske porođajne mase koje se prezentiralo kliničkom slikom respiracijskog zatajenja uslijed respiratornog distres sindroma i perzistentnog pneumotoraksa s pratećim poteškoćama ventilacije. Obzirom na recidivirajuću prirodu bolesti i ograničene kompenzacijske mehanizme nedonoščeta indicirana je torakotomija te verificirana bronhopleuralna fistula gornjeg režnja desnog plućnog krila. Operativnim zahvatom korigirana je prirođena patološka veza između bronha i pleuralne šupljine. Tijek operacije protekao je uredno kao i daljnji postoperativni tijek na odjelu.

Zaključak: Bronhopleuralne fistule su rijetke kongenitalne malformacije i veliki problem kod nedonoščadi. Klinički znak koji je sugerirao na istu je perzistentni i recidivirajući pneumotoraks koji bi bez liječenja rezultirao opstruktivnim šokom i srčanim zastojem uslijed sloma kompenzacijskih mehanizama. Za dobar ishod našeg pacijenta ključan je bio pravodoban dijagnostičko terapijski pristup.

Ključne riječi: perzistentni pneumotoraks, bronhopleuralna fistula, sekundarni pneumotoraks, nedonošče.

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