

Available online at www.sciencedirect.com

ScienceDirect

journal homepage: www.elsevier.com/locate/radcr



Case Report

Diederik P.J. Smeeing, MD, PhD^a,*, Willemijn M. Klein, MD, PhD^b, Edwin F. Dierselhuis, MD, PhD^c, Horst E. Daniels, MD, PhD^d

^a Department of Surgery, Rijnstate Hospital, Arnhem, The Netherlands

^b Department of Medical Imaging, RadboudUMC, Nijmegen, The Netherlands

^c Department of Orthopedic Surgery, RadboudUMC, Nijmegen, The Netherlands

^d Department of Surgery, RadboudUMC, Nijmegen, The Netherlands

ARTICLE INFO

Article history: Received 12 November 2023 Revised 9 March 2024 Accepted 13 March 2024

Keywords: Diaphragmatic hernia Rib osteochondroma Rib exostosis Hereditary multiple exostoses Multiple osteochondroma

ABSTRACT

Diaphragmatic hernia in children is uncommon, especially when not congenital. We present a case of an 11-year old boy with a diaphragmatic hernia caused by a rib osteochondroma. The osteochondroma was surgically removed and the laceration in the diaphragm was repaired. This case shows the importance of being familiar with acquired diaphragmatic hernia in children, to recognize and prevent possible complications in an early stage.

© 2024 The Authors. Published by Elsevier Inc. on behalf of University of Washington. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/)

Introduction

Diaphragmatic hernia in children is uncommon, especially when not congenital. Congenital diaphragmatic hernia (CDH) occurs in 2.3 of every 10,000 births [1]. CDH is caused by incomplete closure of the diaphragm and often identified prenatal or soon after birth. Traumatic diaphragmatic hernia is very rare in the pediatric population and usually accompanied by other severe injuries. Therefore, diagnosis may be delayed [2].

Case description

An 11-year old boy presented to the orthopedic out-patient clinic with progressive pain to the chest and between his scapulae for the past 3 months. He was known with hereditary multiple exostoses/multiple osteochondroma (HME/MO), like his father. He did not have any other medical history. Since multiple osteochondroma can cause bony deformities or leg-length discrepancy in the growing child, he had clinical

* Corresponding author.

https://doi.org/10.1016/j.radcr.2024.03.039

 $^{^{*}}$ Competing Interests: The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

E-mail address: diederiksmeeing@hotmail.com (D.P.J. Smeeing).

^{1930-0433/© 2024} The Authors. Published by Elsevier Inc. on behalf of University of Washington. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/)



Fig. 1 – The 3D reconstruction of the preoperative low dose CT scan, indicating the osteochondromas of the left ribs on the pleural sides.

follow-up on a regular base. The osteochondroma were located at his tibia, fibula, knees, and humeri. These gave minimal symptoms and were without functional limitations. The chest pain with which he now presented was most prominent at nighttime and was sometimes combined with symptoms of dyspnea. The patient was able to point out the maximum pain at the 11th rib on the left side. At physical examination no evident osteochondroma of the rib could be seen or palpated. A low-dose computed tomography (CT) scan of the thorax was made which showed multiple osteochondroma at the ventrolateral pleural side of ribs 2 to 8 on the left side. In addition, pleural thickening at the site of the osteochondromas, ground-glass opacity and pleural fluid were seen. The CT scan was suggestive for irritative pleuritis caused by the rib osteochondromas. Due to the progressive character of the pain he was planned for resection of the largest osteochondromas.

Fifteen days after initial presentation resection was performed of the largest osteochondromas which were located at rib 3 to 6 (Figs. 1 and 2). As a result of the profound relation of the pleura to the rib osteochondromas a small lesion of the pleura occurred during surgery for which a water sealed chest tube was placed. Postoperative the patient underwent chest Xray because of thoracic pain and tachypnea. The chest X-ray showed a pneumothorax, an in situ chest tube and a poorly demarcated left dome of the diaphragm (Fig. 3). After pain man-



Fig. 2 - One of the excised rib osteochondroma.



Fig. 3 – The postoperative chest x-ray shows a pneumothorax on the left side, with a chest tube and a poorly demarcated left dome of the diaphragm, suggesting postoperative pleural fluid, atelectasis and consolidation.



Fig. 4 – The postoperative Contrast-enhanced CT scan (coronal view) shows a ventrolateral diaphragmatic lesion in combination with a partial herniation of colon and indurated mesenterial fat.



Fig. 5 – Diaphragmatic defect visible through a lateral mini-thoracotomy.

agement the symptoms of the patient slowly improved. Considering the combination of the postoperative clinical symptoms of the patient and the chest X-ray, the pre-operative lowdose computed tomography of the chest was reassessed after which the suspicion of a small left-sided diaphragmatic hernia was raised. After multidisciplinary consultation (pediatric surgery, pediatrics, orthopedic surgery, and radiology) the decision was made to perform a new thoracic CT-scan to confirm the suspicion and its current status. It showed a ventrolateral diaphragmatic laceration close to a rib osteochondroma in combination with a partial herniation of colon and indurated mesenterial fat (Fig. 4). The patient was without abdominal symptoms and did not show any signs of peritoneal stimulation. An urgent reoperation was performed in which a sharp rib osteochondroma of approximately 2 cm in length was removed through a lateral mini-thoracotomy (Figs. 5 and 6). This osteochondroma of rib 8 was suspected to have caused the diaphragmatic laceration as it's sharp aspect slided against the diaphragm at expiration. Through the same lateral mini-thoracotomy the colon and omentum were inspected for signs of ischemia or lesions and subsequently repositioned. The defect in the diaphragm of about 2 cm was closed using standing sutures. The chest tube could be removed 1 day after the operation. The pathology results confirmed the diagnosis of osteochondroma. The patient recovered quickly and was discharged from the hospital 3 days postoperative. After two years the patient is still fine without remaining thoracic symptoms or recurrence.

Discussion

Diaphragmatic hernia is an infrequent cause for pathology in children [1]. A congenital diaphragmatic hernia is often



Fig. 6 – Removed sharp rib osteochondroma of approximately 2 cm on a surgical gauze.

diagnosed prenatal or shortly after birth due to the severity of symptoms; although there are asymptomatic cases. Traumatic or iatrogenic diaphragmatic hernia often presents later in life and have a sudden onset. A traumatic diaphragmatic hernia is often accompanied by both abdominal and thoracic injuries. They can be caused by blunt and penetrating trauma [3]. Iatrogenic injuries to the diaphragm are rare, but are described after surgery like esophagectomy, gastrectomy, laparoscopic cholecystectomy, or nephrectomy [4-9]. In this case report, a rib osteochondroma caused the diaphragmatic laceration. Rib osteochondroma can occur in patients with hereditary multiple osteochondromas (HMO), also called hereditary multiple exostoses (HME). The prevalence of HMO is estimated at 1 in 50,000 people [10]. The osteochondromas can occur all over the body, but predominantly arise at the long bones of the extremities [10]. Rib osteochondromas have been described to cause: hemothorax, pneumothorax, and thickening of pericardium [11]. A first symptom could also be a "spontaneous" pneumo- or hemothorax [12]. Spontaneous pneumo- or hemothorax are uncommon and therefore underlying pathology should always be suspected and imaging should be performed. Delayed diagnosis of a diaphragmatic hernia can cause worsening of the patient's condition due to herniation and strangulation of abdominal organs. Therefore, early diagnosis of an injury to the diaphragm is essential, for which a CT scan is advised.

This case report highlights the importance to be aware of acquired diaphragmatic hernia in young children, which can be a complication of a rib osteochondroma.

Patient consent

Hereby I declare that written informed consent for publication of the case was obtained from the patient and both parents.

REFERENCES

- Paoletti M, Raffler G, Gaffi MS, Antounians L, Lauriti G, Zani A. Prevalence and risk factors for congenital diaphragmatic hernia: a global view. J Pediatr Surg 2020;55(11):2297–307.
- [2] AH Al-Salem. Traumatic diaphragmatic hernia in children. Pediatr Surg Int 2012;28(7):687–91.
- [3] Tokgöz S, Akkoca M, Uçar Y, Yilmaz KB, Sevim Ö, Gündoğan G. Factors affecting mortality in traumatic diaphragm ruptures. Ulus Travma Acil Cerrahi Derg 2019;25(6):567–74.
- [4] Celia A, Del Biondo D, Zaccolini G, Breda G. Iatrogenic diaphragmatic lesion: laparoscopic repair. Minerva Urol Nefrol 2010;62(3):327–30.
- [5] Suh Y, Lee JH, Jeon H, Kim D, Kim W. Late onset iatrogenic diaphragmatic hernia after laparoscopy-assisted total gastrectomy for gastric cancer. J Gastric Cancer 2012;12(1):49.
- [6] Bouchagier K, Solakis E, Klimopoulos S, Demesticha T, Filippou D, Skandalakis P. A rare case of iatrogenic diaphragm defect following laparoscopic cholecystectomy presented as acute respiratory distress syndrome. Case Rep Surg 2018;2018:4165842.
- [7] Sasaki M, Takahashi T, Funaki S, Tanaka K, Miyazaki Y, Ose N, et al. A case of diaphragmatic hernia incarceration after a heart transplant operation. Asian J Endosc Surg 2021;14(1):116–19.
- [8] Saito T, Yasui K, Kurahashi S, Komaya K, Ishiguro S, Arikawa T, et al. Intrapericardial diaphragmatic hernia into the pericardium after esophagectomy: a case report. Surg Case Rep 2018;4(1):94.
- [9] Pan SB, Zhang JB, Zhao BQ, Chai Y. Delayed iatrogenic diaphragmatic hernia after thoracoscopic lobectomy. J Thorac Dis 2016;8(6):E399–402.
- [10] Bovée JVMG. Multiple osteochondromas. Orphanet J Rare Dis 2008;3:3.
- [11] Assefa D, Murphy RC, Bergman K, Atlas AB. Three faces of costal exostoses: case series and review of literature. Pediatr Emerg Care 2011;27(12):1188–91.
- [12] Khosla A, Parry RL. Costal osteochondroma causing pneumothorax in an adolescent: a case report and review of the literature. J Pediatr Surg 2010;45(11):2250–3.