International Journal of Surgery Case Reports 99 (2022) 107714

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

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

Case report

Case report: A rare case of congenital non-metastatic low-grade fibrosarcoma of the pleura in a 6-month-old infant manifested as pneumonia

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ARTICLE INFO

Keywords:
Case report
Congenital fibrosarcoma
Spindle-cell sarcoma
Pleural sarcoma
Rare childhood tumors

ABSTRACT

Introduction and importance: Congenital Infantile fibrosarcoma is a rare tumor in children and accounts for only 10 % of various malignant tumors in this age group. Manifestations vary according to the site of occurrence. Symptoms of the tumor located in unusual places can be misleading and obscure the actual diagnosis, which in turn may waste precious effort and time until the correct diagnosis is established. Infant malignancies should be considered to reduce the morbidity and mortality associated with this pathology.

Case presentation: We present the case of a 6-month-old infant, who was admitted to our hospital with a one-month history of high fever and dry cough, with no improvement in symptoms after treatment with antibiotics. Computed tomography showed a heterogeneous mass in the pleural cavity compressing the lung tissue, in addition to bilateral mild pleural effusion. Thoracotomy was indicated and the tumor was completely resected.

Clinical discussion: Histopathological and Immunohistochemical approach is crucial because this type of tumor can overlap with many soft tissue sarcomas. After searching in medical literature, no published evidence of a similar case was found, and thus we managed the patient empirically, depending on the usual approach for congenital fibrosarcoma. Complete surgical resection is the golden standard of treatment, followed by chemotherapy depending on pathological findings.

Conclusion: Clinical awareness is important in any unresponsive pneumonia and malignancies should be taken into consideration.

1. Introduction

Solid tumors are considered rare in the newborn or the neonatal period and are often benign. The presence of such tumors represents only 2 % of different malignant tumors in childhood. Soft tissue sarcomas are a group of tumors that differentiate from mesenchymal cells and this means that they can differentiate into muscular or lipid tissue as well as fibrous tissue and other tissues. Fibrosarcoma can be classified into two categories during this age phase: congenital Infantile fibrosarcoma (CIFS) and adult-type fibrosarcoma, which have similar features but vary by outcomes. In childhood, CIFS is rare and makes up only 10 % of various malignant tumors in children [1]. We often see congenital fibrosarcoma in the limbs and, less commonly, in the trunk, head, and neck. The presence of congenital fibrosarcoma in the chest cavity is unusual as the medical literature has never mentioned a similar case.

The basic and final diagnosis is made by the integration of both clinical and radiological findings in addition to histopathology [2].

Both fibrosarcoma patterns are similar but we can clinically distinguish between them (the adult type is usually in children aged 10 to 15). Histological results show that the tumor consists of spindle-shaped fibroblasts. A recurring translocation (t(12;15)) involving the ETV6 and NTRK3 genes has been documented in CIF, but not in the 'adult type'. This model of transportation may be associated with a better prognosis. [1].

The work has been reported in line with the SCARE criteria and the

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https://doi.org/10.1016/j.ijscr.2022.107714
Received 18 August 2022; Received in revised form 25 September 2022; Accepted 26 September 2022
Available online 28 September 2022

revised 2020 SCARE guidelines [3].

2. Presentation of case

2.1. Patient information

We demonstrate the case of a 6-month-old male who was born via elective caesarian; presented to the Emergency Department (ED) with fever, dry cough, tachypnea, and progressive shortness of breath. The story started 1 month before admission with a complaint of high fever and cough. He was taken to the emergency department in a rural hospital, where he was diagnosed with acute pneumonia and treated with intravenous (IV) antibiotics for 13 days in the hospital. Symptoms were relieved temporarily and then recurred 5 days after discharge. As a result, he was readmitted to the hospital and treated for bilateral pneumonia for 17 days with only slight improvement. Therefore, he was referred to our university hospital for a thorough study of the case. On admission, the patient had tachycardia and tachypnea with mild dehydration. No genitourinary or gastrointestinal symptoms were reported. Family, drug, and allergic histories were negative.

2.2. Clinical findings

Clinical examination revealed tachycardia and tachypnea with prolonged exhalation and bilateral soft rales upon auscultation, with right basal dullness in chest percussion.

Laboratory investigations revealed low hemoglobin value (7.1 Mg /dl), low MCV (66 fl) with leukocytosis (WBC: 12.7/μL) and elevated CRP (77 mg\L) but otherwise, within normal values.

2.3. Diagnostic assessment

Chest X-Ray showed bilateral pulmonary infiltrates, left hyperventilation, right middle lobe atelectasis, and right pleural effusion.

CT scan of the chest (Fig. 1) showed a heterogeneous mass in the posterior part of the right lower lobe, that measures (75 × 64 × 43 mm) and compress the remaining pulmonary tissue.

Initial treatment involved intravenous fluid resuscitation, wide spectrum antibiotics, complete laboratory investigations panel including sampling and crossmatch of blood group for blood transfusion to reclaim the patient's hemoglobin levels.

Tumor markers were remanded: Alpha fetoprotein (AFP) = 65 (N > 10) – Neuron specific enolase (NSE) = 39.17 (N > 17).

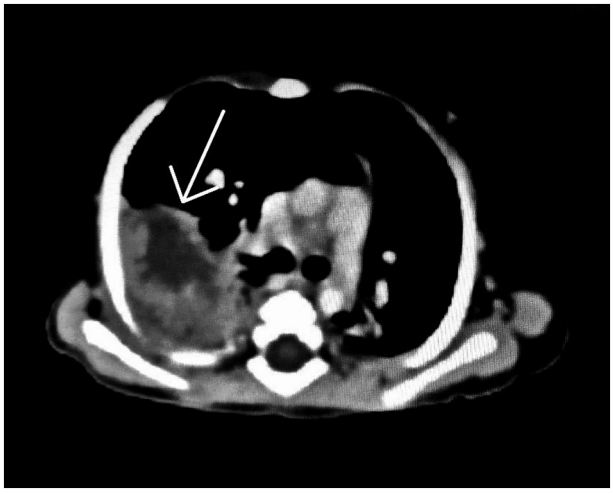

Fig. 1. CT scan/cross sectional view of the Chest showing a heterogeneous mass in the posterior part of the right lower lobe (white arrow) that measures (75 × 64 × 43 mm) with a compressive effect on the remain of pulmonary tissue.

To make sure that there were no distant metastases before surgery, a head /chest/ abdomen/ limbs MSCT scan was performed. The results were discussed in an MTD meeting and it was confirmed that there were no distant metastases.

No predisposing risk factors were identified, as the mother did not report any exposure to radiation or any other carcinogens during pregnancy. In addition, no family history of cancer has been documented.

2.4. Therapeutic intervention

Open surgery was indicated based on the given clinical picture. The procedure was performed at our tertiary university hospital by a sixth-year senior Pediatric Surgery resident with six years of surgical experience and by a Pediatric Surgery specialist with 15 years of Pediatric Surgery experience. The procedure was done under general anaesthesia with no anesthetic complications. Right posterolateral thoracotomy revealed a (10 × 8 × 4 cm) solid pleural mass located posterolateral to the right lower lobe of the right lung (Fig. 2A–B). the mass was fully resected with several lzmph nodes from the right hilum and sent to histological examination.

Preliminary histopathology revealed spindle-cell neoplasm consistent with low-grade fibrosarcoma (Fig. 3) and mild nonspecific pleuritis. All of the isolated lymph nodes were free of invasion by neoplastic cells.

Immunohistochemistry confirmed the diagnosis with (VIM positive, S100 negative, Desmin negative, Ki 67 < 3 %) (Fig. 4A–D).

The patient had an uneventful postoperative recovery and therefor, was discharged after 7 days from the operation. He has been followed up as an outpatient for 10 months following his operation and referred to a specialized oncology tertiary university hospital, where he received adjuvant chemotherapy of Vincristine, actinomycin D, and cyclophosphamide (VAC). We could find no published evidence of a similar case

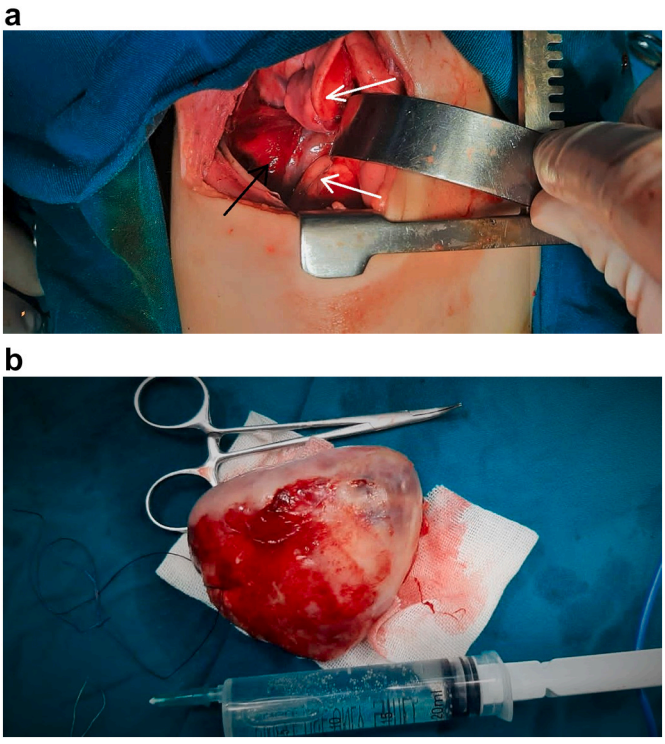

Fig. 2. A: Intraoperative image prior to tumor excision. Black Arrow points toward the solid pleural mass that is located posterolateral to the right lower lobe of the right lung (white arrows).
B: intraoperative image post-resection of the tumor, represent the excise-dpleural tumor.

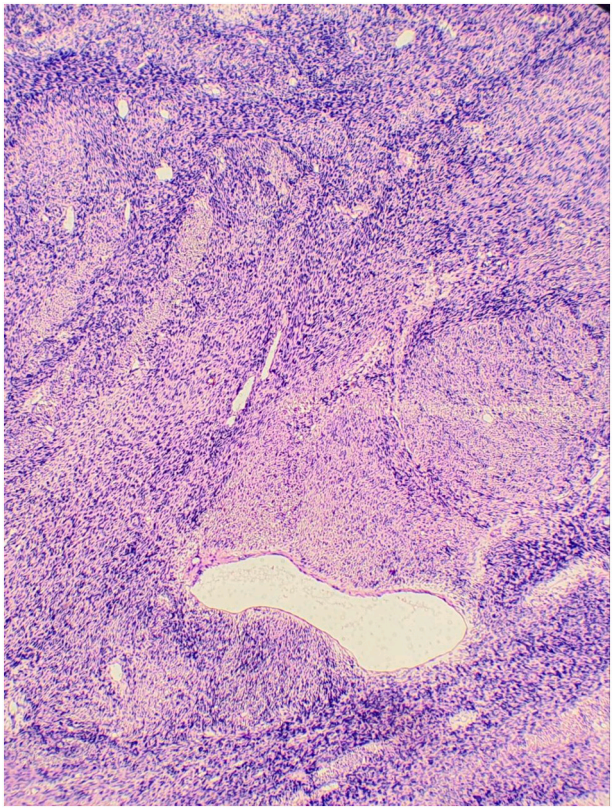

Fig. 3. H&E stain showing spindle cell neoplasm consistent with low grade fibrosarcoma.

and therefore managed the patient empirically, using a standard sarcoma chemotherapy protocol. In addition, he registered regularly scheduled visits with the pediatric surgery clinic and the oncology clinic to monitor his disease progression and evaluate his response to treatment. The evaluation included physical examination, laboratory tests, and several postoperative CT scans of the chest. The postoperative CT scans of the chest revealed the following: the chest was clear and free of any reactive lymphadenopathy, slight atelectasis in the apical segment of the right inferior lobe. (Fig. 5).

3. Discussion

Congenital infantile fibrosarcoma (CIFS) is a rare tumor that is often seen after birth and evolves during the early years of life. CIFS represents about 1 % of tumors observed during childhood and 5 to 10 % of soft tissue in infants younger than one-year-old [4–9]. These tumors occur in about five cases per million infants [10–12]. One-third of patients manifest in the perinatal period [8,10]. About 50 % of these tumors arise before the age of 3 months [14]. A second peak of the tumor's onset was recorded at the age of 10 to 15 years [11,13]. In addition, these tumors were reported prenatally by diagnosing via ultrasound or MRI [15]. The incidence rate of these tumors in males is higher than in females and is estimated at (1/3 to 1/4). Only about 60 cases of these tumors have been documented as congenital in the past 40 years [16,17]. Distant limbs are the most common site of occurrence [18–20], while the head, neck, and trunk are rare sites of manifestation [21], as well as the chest wall [18]. By reviewing the medical literature, we were unable to find any case of congenital infantile sarcoma that had been documented to occur at the expense of the pleura. CIFS is divided into two subtypes in children: Desmoplastic and medullary types. The desmoplastic-type tends to be locally aggressive and has histopathological similarities to adult-type fibrosarcoma. While the Medullary-type is less aggressive behavior and have a benign clinical path [22]. The location of the tumor is the main determinant of the clinical presentation rather than the tumor's subtype. When the tumor is located in the chest wall, paravertebrally, or in the posterior mediastinum, then the typical symptom would be a pain in the chest, shoulder, neck, or back, whereas cough, dyspnea, and hemoptysis dominate when the tumor is centered on the airways. On the other hand, tumors occupying the pleural cavity, such as the primary pleural masses or peripheral intrapulmonary tumors, induce pleural effusion. In our case, because of the scarcity of the presence of this tumor in the chest cavity in children, we didn't find specific documented symptoms related to it. Despite similar histopathological features and malignant appearance to adult-type fibrosarcoma, CIFS is considered a different clinical entity with a good prognosis and different cytogenetic specifications [4,8]. CIFS tends to behave benignly and rarely metastasizes [23,24], as metastases are found in <10 % of patients under the age of 5, while >50 % of patients over the age of 10 tend to have metastases [9,25,26]. The metastases usually locate in lungs, bones and sometimes lymphatic nodes [18,27–31]. Localized recurrence is common and makes up about 20 to 40 % of cases [19,27,29–31]. This tumor is classified a low-grade myofibroblastic Non-Rhabdomyosarcoma Soft-Tissue Sarcoma (NRSTS) [4,7,9,25,32]. When examined under a microscope, this tumor consists of spindle cells arranged in bundles and fascicles showing a herringbone pattern.

Immunohistochemistry is a significant assistant in the diagnosis of soft tissue neoplasms. Because of the morphological interference of soft tissue tumors with several neoplasms including infantile fibromatosis and myofibromatosis. CIFS has strong positive Vimentin, smooth muscle actin (SMA), and about 25 % of Desmin markers, while CD34 and S-100 protein and myoglobin are generally negative [4,5].

Cytogenetic analysis with RT- PCR and FISH may show some specific abnormalities for CIFS and some non-specific for congenital infantile myofibromatosis. Precisely, CIFS seems to include a translocation t (12;15) (p13;q25) resulting in ETV6 – NTRK3 gene fusion. This translocation was seen in another tumor of infancy which is congenital mesoblastic nephroma. In addition, in CIFS, some polysomies were detected in particular chromosomes (trisomies of chromosomes 11, 8, 20) [4,8,33]. The diagnosis of CIFS is established based on the exclusion of the other tumors according to immunohistochemistry results as well as the classic “herring-bone” pattern.

The findings on plain X-ray are nonspecific and the soft tissue tumor may not appear clearly. Ultrasonography is a prenatal survey where it can sometimes show soft tissue tumors before birth and can be used to guide biopsy when the tumors are superficial [34,35]. CT scans can detect tumors with bony involvement. The favorable imaging modality to evaluate the tumor, its extension, and its relationship to adjacent tissue is magnetic resonance (MRI) [4,31]. Regarding treatment, wide surgical resection (WSE) is the initial treatment and is often sufficient in chest wall lesions [18,20], and the sufficiency of complete resection is the most important factor affecting patient survival, with a survival rate reaching 90 % in long-term studies. Neoadjuvant chemotherapy hasn't been determined officially but is often applied before surgery to improve the possibility of total removal of the tumor. For older children, chemotherapy is used to reduce the likelihood of metastases. When the edges of the surgical removal of the tumor contain tumor cells microscopically or when the complete removal is not possible, the application of postoperative chemotherapy is considered, the chemotherapy regime is usually (vincristine, cyclophosphamide, and actinomycin D) [36]. On the other hand, secondary malignant tumors should be taken into consideration when applying adjuvant chemotherapy at this age [8,37]. Moreover, Radiotherapy is used only as a palliative treatment due to developmental disorders that could be caused by it [8,37]. Although pleural tumors are uncommon, there is a vast field of differential diagnosis including soft tissue sarcomas like spindle cell rhabdomyosarcoma, synovial sarcoma, and infantile hemangiopericytoma, and the cellular form of childhood fibromatosis must be ruled out, due to their invasive behavior [28].

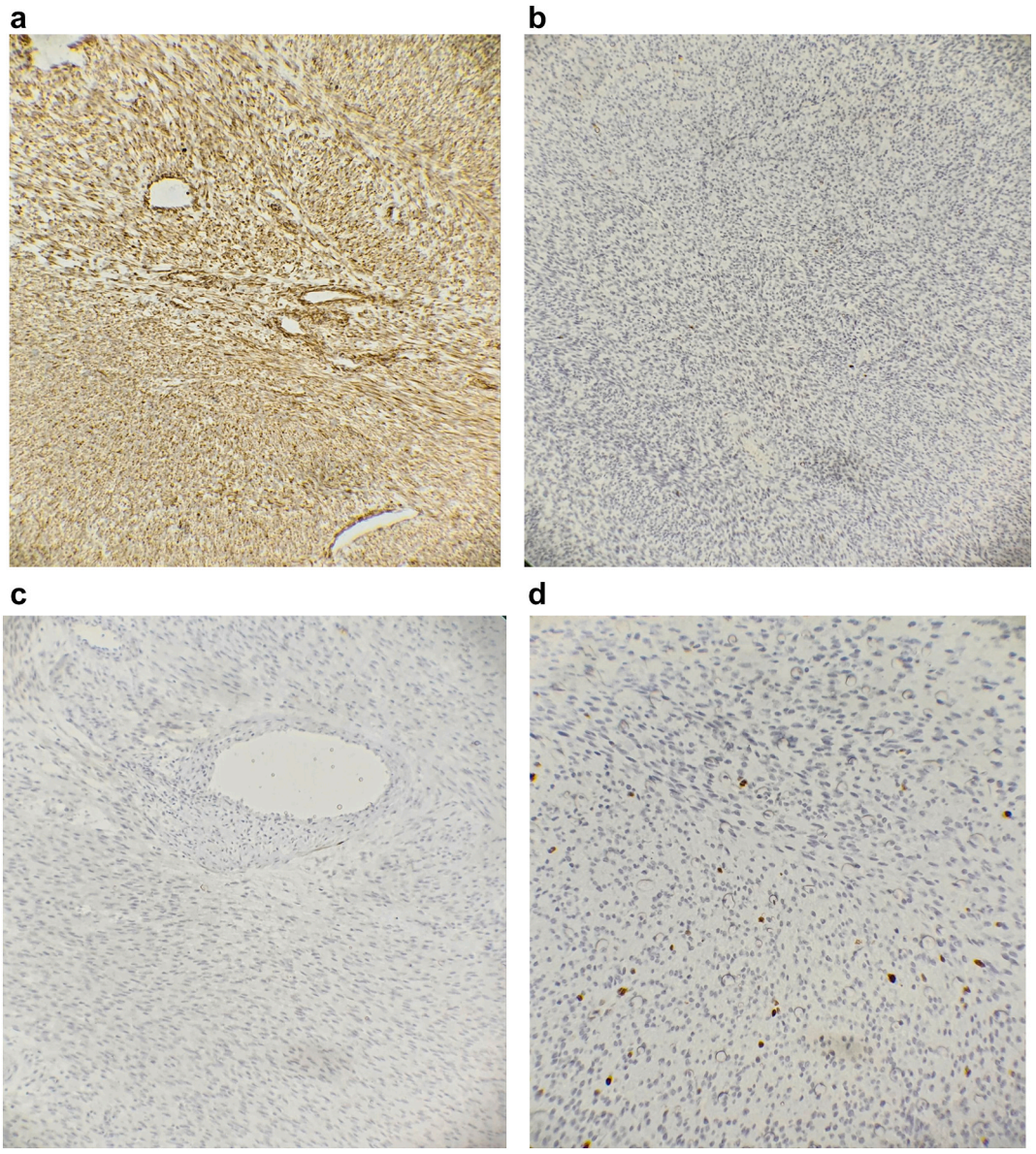

Fig. 4. A: immunohistochemistry stain revealed vimentin marker to be positive.
B: immunohistochemistry stain revealed S100 marker to be negative.
C: immunohistochemistry stain revealed Desmin marker to be negative.
D: immunohistochemistry stain revealed ki 67 marker to be >3 %.

4. Conclusion

Fibrosarcoma of pleura is rare in infants, and requires a high index of suspicion to diagnose and a multidisciplinary team for treatment, which is surgical resection in suitable cases followed by chemotherapy.

Abbreviations

CIFS	Congenital infantile fibrosarcoma
CT	computed Tomography
IV	intravenous

Ethics approval and consent to participate

Institutional review board approval is not required for deidentified single case reports or histories based on institutional policies.

Consent of patient

Written informed consent was obtained from the patient's parents for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Availability of data and materials

The datasets generated during and/or analyzed during the current study are not publicly available because the Data were obtained from the hospital computer-based in-house system. Data are available from the corresponding author upon reasonable request.

Provenance and peer review

Not commissioned, externally peer-reviewed.

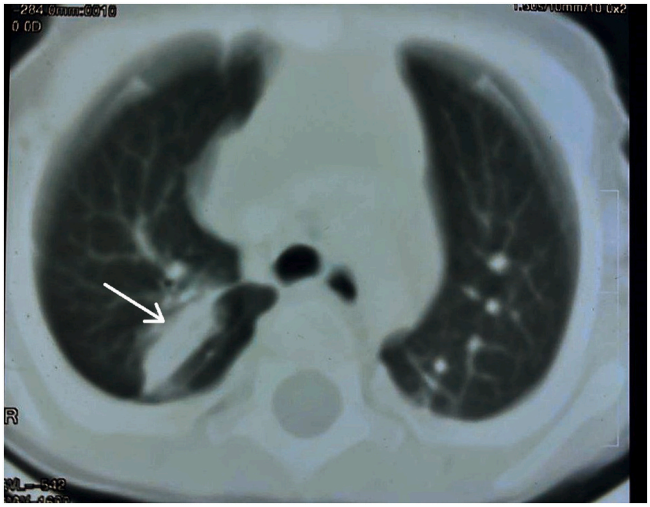

Fig. 5. Post- Operation CT scan/cross sectional view of the Chest showing clear right lung field except a slight atelectasis in apical segment of the right inferior lobe (arrow).

Sources of funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Research registration

N/A.

Guarantor

Khaled Alomar

CRediT authorship contribution statement

RA, KA, RSA: Who wrote, original drafted, edited, visualized, validated, literature reviewed the manuscript.

FAS: General Surgery senior resident who was the first assistant in the surgery. Supervision, project administration, and review of the manuscript.

IEAS: pediatric Surgery Consultant, who performed and supervised the operation.

OH: MD, PhD, head of hematology and oncology department in university pediatrics hospital, supervision and review of the manuscript.

KA: Conceptualization, resources, and the corresponding author who submitted the paper for publication.

All authors read and approved the final manuscript.

Declaration of competing interest

The authors declare that they have no competing interests.

Acknowledgements

- Alshaty Histopathology and Immunohistochemistry Laboratory, Damascus, Syria.
- Hematology and oncology department in university pediatrics hospital.

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