Mesenchymal Hamartoma of the Chest Wall in a Newborn: A Case Report Study

Abstract

Mesenchymal hamartoma of the chest wall is an extremely rare benign lesion arising from one or more ribs in neonates. Its incidence is estimated to be <1 case in a million in general population. It usually presents in the form of chest wall mass. Respiratory distress may be existed resulting from compression of the airways and lungs. Herein, we reported a 1-day-old neonate with respiratory distress and mild chest wall deformity. Imaging studies revealed an extrapleural solid cystic lesion in the middle zone of the left hemithorax. When her respiratory distress was managed, she underwent left thoracotomy, resection of chest wall tumor, and thoracoplasty with titanium mesh. Six months of follow-up revealed no evidence of recurrence. The findings of this case report yielded that mesenchymal hamartoma is a benign lesion presenting with aggressive clinical, radiological, and histopathological characteristics that can be mistaken for malignancy.

Keywords: *Benign lesion, mesenchymal hamartoma, neonate, thoracoplasty, thoracotomy*

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Introduction

Chest wall tumors are very rare in children.[1] Mesenchymal hamartoma of the chest wall is an unusual extrapleural tumor originating from the ribs.[2] In addition, mesenchymal hamartoma accounts for only 0.03% of primary bone tumors.[3,4] It usually presents at birth or during early infancy.[5] The sufferers may be presented with respiratory distress or an asymptomatic mass arising from the ribs.[6] Despite its aggressive clinical, radiographic, and histopathological characteristics, mesenchymal hamartoma has a benign nature. On the other hand, its differentiation from malignancy can be challenging.[7] In the current investigation, we reported mesenchymal hamartoma of the chest wall in a 1-day-old neonate presented with respiratory distress and mild chest wall deformity.

Case Report

A 1-day-old neonate was admitted to the neonatal intensive care unit (NICU) of Shahid Sadoughi Hospital, Yazd, Iran, because of respiratory distress and tachypnea. The infant weighed 3100 g at birth and was delivered at 39 weeks and 4 days of gestation. At the time of her admission to NICU, her vital signs were as follows: pulse rate = 150 beats/minute, respiratory rate = 65/minute, temperature = 36.8°C, and O2 saturation=85%.

Physical examination revealed marked tachypnea, subcostal and intercostal retractions, and mild chest wall deformity. Her cardiac auscultation was normal. Breath sounds on the left lower side of the thorax were markedly decreased. Sucking, grasping, and parachute reflexes were all normal.

The results of blood gas analysis revealed that her pH = 7.20, HCO_3 = 25.4, PO_2 = 42, and PCO_2 = 62. The findings of her laboratory tests were as follows: white blood cell = 16,700/mm^3, hemoglobin = 17.4 g/dL, platelets = 214,000/μL, Na = 140 mEq/L, K = 4.8 mEq/L, Ca = 9.5 mg/dL, BUN = 19, Cr = 0.9 mg/dL, prothrombin time = 14.4 s, partial thromboplastin time = 34.1 s, INR = 1.19, lactate dehydrogenase = 36 U/L, BhcG = 1.05, and αFP >300 ng/mL. Prenatal ultrasound examination reported type II congenital cystic adenomatoid malformation, leading to shift of her heart to the right side at 36 weeks' gestation. Chest X-ray showed lytic and sclerotic lesions in the 4th, 5th, and 6th left ribs [Figure 1].

Computed tomography (CT) of the thorax demonstrated an extrapleural solid cystic lesion in the middle zone of left hemithorax

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measuring 58 mm × 53 mm, which was associated with expansion and destruction of the 4th, 5th, 6th, and 7th left ribs. The findings of CT demonstrated shifting of her heart and mediastinum to the right side.

Following clinical stabilization of the patient, the mass was excised through operation. The baby underwent left thoracotomy, chest wall tumor resection, and thoracoplasty with titanium mesh. After skin preparation and draping, the chest wall (skin, muscles) was opened by oblique incision over the 5th rib under general anesthesia in the lateral position. The mass was firm and fixed to the 5th, 6th, and 7th ribs. Thorax cage was opened over the 4th intercostal space. The mass extended out of the one-third anterior of ribs to the vertebral column. The lung was free from any tumors. The tumor was totally excided with resection of four ribs. The chest wall defect was reconstituted with titanium mesh. The chest tube was also inserted [Figure 2].

Histopathological examination of hematoxylin and eosin-stained glass slides revealed tumoral lesion composed of spindle cells arranging in fascicles and whorled pattern with mitotic figures. Large blood vessel channels were observed, which were lined by multinucleated giant cells and mixed with spindle cells. Numerous multinucleated giant cells and cells with osteoid formation were also discovered [Figure 3]. These findings were consistent with diagnosis of mesenchymal hamartoma.

Immunohistochemical studies were done and cells showed positive reaction pattern to the S100, vimentin, and CD68. However, epithelial membrane antigen, myogenin, CD34, desmin, and pan-cytokeratin were all negative.

Discussion

Mesenchymal hamartoma of the chest wall is an extremely rare benign lesion in neonates and pediatrics.[8] Its incidence is estimated to be <1 in a million in general population. Mesenchymal hamartoma usually occurs unilaterally, and there are few case reports in this regard introducing bilateral lesions.[3]

Mesenchymal hamartoma derives from rib cage, and it is a combination of benign proliferation of skeletal tissues, particularly cartilaginous components and hemorrhagic cavities.[9] In addition to the visible chest wall mass at birth, respiratory distress may also be present in this disease depending upon the size of extrapleural mass. Displacement of the heart and lungs can be also associated with the incidence of mesenchymal hamartoma. The course of the disease is usually benign, but it can be fatal in some cases.[10,11] In the current study, we introduced a 1-day-old patient with respiratory distress and mild unilateral chest wall deformity.

Since imaging studies are helpful for the diagnosis of mesenchymal hamartoma and CT scan generally is able to demonstrate intralesional and multifocal calcification,[12] we used this technique for the diagnosis. CT scan findings showed an extrapleural solid cystic lesion in the middle zone of left hemithorax associated with expansion and destruction of the 4th, 5th, 6th, and 7th left ribs.

Mesenchymal hamartoma may be misdiagnosed with enchondroma, fibrous hamartoma, malignant mesenchymoma, and aneurysmal bone cyst.[13] We can achieve definitive diagnosis only by resorting to histopathological examinations.[14] Microscopically, it is composed of mesenchymal cells, osteoclastic giant cells, hyaline cartilage, and aneurysmal bone cyst-like changes.[15] In our case, tumoral lesion involved spindle cells that were arranged in fascicles and whorled pattern with mitotic figures. Large blood vessel channels were also observed, which were lined by multinucleated giant cells and mixed with spindle cells. Numerous multinucleated giant cells and cells with osteoid formation were also discovered. These findings were consistent with the diagnosis of mesenchymal hamartoma in our case.

Surgical resection is the preferred strategy for patients suffering from respiratory distress.[16] However, conservative

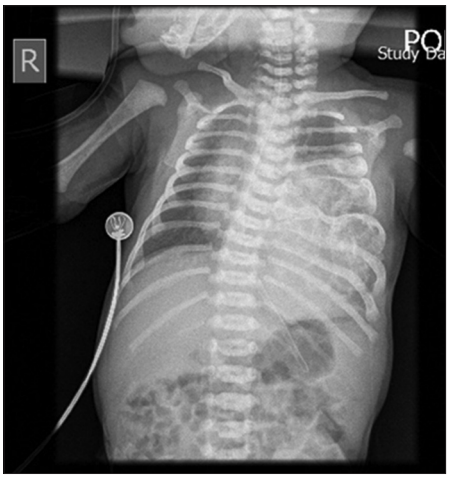

Figure 1: Chest X-ray showing lytic and sclerotic lesions in left 4th, 5th, and 6th ribs

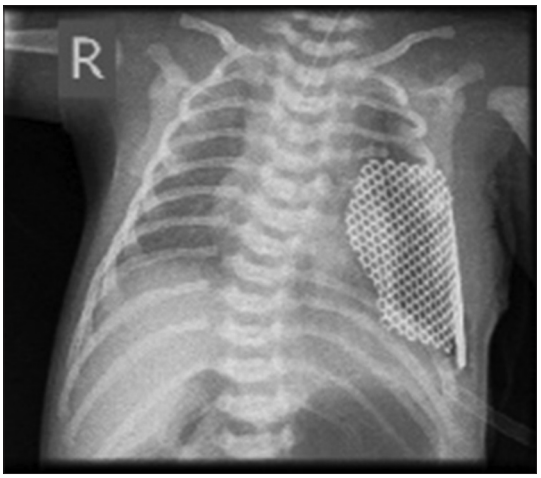

Figure 2: In the postoperative chest X-ray, reconstitution of chest wall with titanium mesh is notable

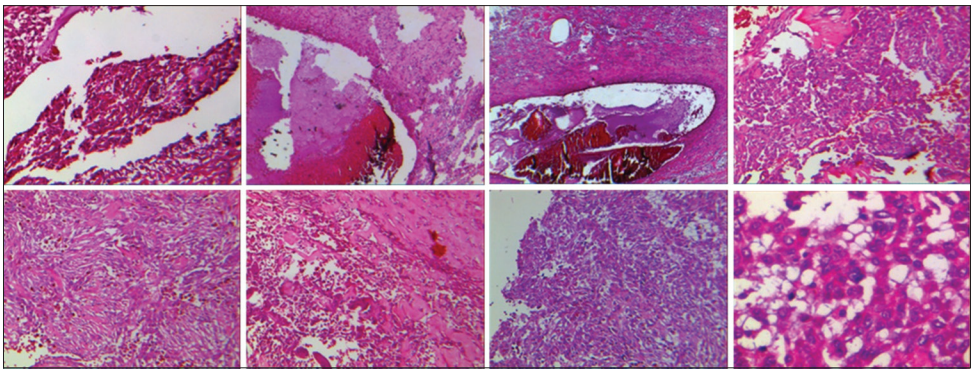

Figure 3: Microscopic examination showing tumoral lesion composed of spindle cells that arranged in fascicles and whorled pattern with mitotic figures. Large blood vessel channels were present that lined by multinucleated giant cells and mixed with spindle cells. Numerous multinucleated giant cells and osteoid formation were also present

treatment may be considered for some cases. After being diagnosed with mesenchymal hamartoma of the chest wall, our patient underwent left thoracotomy, resection of chest wall tumor, and thoracoplasty with titanium mesh. Her 6 months of follow-up revealed no sign of recurrence.

Conclusion

We introduced chest wall mesenchymal hamartoma in a newborn presented with respiratory distress. We came to this finding that mesenchymal hamartoma usually has a benign course, although it may have aggressive radiological and histological features.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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