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Chest wall osteochondroma resection with biologic acellular bovine dermal mesh reconstruction in pediatric hereditary multiple exostoses: A case report and review of literature

Alshehri A. Biologic mesh reconstruction in pediatric HME

Abstract

BACKGROUND

Hereditary multiple exostoses (HME) are a rare genetic disorder characterized by the growth of multiple osteochondromas affecting primarily long bones. Chest wall lesions may represent a challenge, particularly in pediatric patients. Pain is a common manifestation. However, life-threatening complications can result from direct involvement of adjacent structures. Surgical resection with appropriate reconstruction is often required.

CASE SUMMARY

A 5-year-old male who was diagnosed with hereditary multiple exostoses presented with significant pain from a large growing chest wall exostosis lesion. After appropriate preoperative investigations, he underwent surgical resection with reconstruction of his chest wall using biologic bovine dermal matrix mesh.

CONCLUSION

Resection of chest wall lesions in children represents a challenge. Preoperative planning to determine the appropriate reconstruction strategy is essential.

Key Words: Hereditary multiple exostoses; Chest wall neoplasm; Chest wall reconstruction; Biologic mesh; Pediatric; Case report

INTRODUCTION

Exostosis, also known as osteochondroma, is a benign osseous outgrowth that can occur either sporadically or as a manifestation of a genetic disorder known as hereditary multiple exostoses (HME). The exact etiology of the disease is unknown. However, it is hypothesized the lesions originate from separation of a portion of the epiphyseal growth plate cartilage from the main epiphysis[1-4]. Although these lesions are usually well tolerated, exostoses might cause significant pain and disabilities. More serious complications have been reported including impingement on neural or vascular structures, joint motion limitation, deformities, limb discrepancy, or malignant transformation[5]. Chest wall osteochondromas can present with life-threatening complications such as pneumothorax, hemothorax, pericardial effusion, or diaphragmatic injury[6-13]. Resection is typically recommended for symptomatic exostoses, and reconstruction may be required depending on several factors[5,14]. A wide range of prosthetic material including biologic meshes have been used to reconstruct the chest wall in pediatric and adult patients[15-23].

In this case report, a 5-year-old male presented with a painful large chest wall exostosis that was resected. Chest wall reconstruction using a biologic mesh derived from acellular bovine dermis was performed as well.

CASE PRESENTATION

Chief complaints

A 5-year-old male presented to our pediatric surgery clinic with a painful mass in the chest wall.

History of present illness

The patient had been diagnosed previously with HME at the age of 1 year. He had multiple bony lesions affecting his chest wall and upper and lower extremities. Despite occasional pain over the bony lesions, his overall functional status was excellent. Since the age of 3 years, his parents noticed a bony lesion over the right side of his chest that was growing. It started to cause significant pain and affected his sleep, right arm movement, and overall ability to attend his school.

History of past illness

Besides diagnosis of HME, the patient had no other illnesses nor any known allergies.

Personal and family history

The patient had a strong family history of HME, affecting both his brother and uncle.

Physical examination

The patient had normal vital signs. His chest examination revealed a 10 cm irregular, hard mass over the right anterior chest centered along the anterior axillary line. The overlying skin was normal (Figure 1). Another 2 cm lesion was felt below the main lesion and close to the right costal margin. A third small lesion was present on the left side of his chest, which was located at the costal margin. Several small bony lesions were felt over his upper and lower extremities close to his elbow and knee joints.

Laboratory examinations

Blood analysis revealed the following: White blood cell count, 12.5 × 10^9/L (reference range: 5.0-15.5 × 10^9/L); red blood cell count, 3.5 × 10^{12}/L (reference range: 3.9-5.0 × 10^{12}/L); hemoglobin, 114 g/L (reference range: 110-138 g/L); and platelet count, 284 × 10^9/L (reference range: 150-350 × 10^9/L). Biochemical analysis revealed normal levels of total protein (58 g/L), albumin (34 g/L), creatinine (28 μmol/L), and urea nitrogen (4.1 mol/L).

Imaging examinations

Chest X-ray showed a distorted right middle of the chest. An enlarged osseous/cartilaginous lobulated exophytic lesion compatible with known exostosis demonstrated a dense matrix compatible with cartilaginous matrix. The heart was not enlarged, and the lungs were otherwise clear (Figure 2).

Computed tomography of the chest revealed large exostosis arising from the anterior portion of the right fourth rib extending to the costochondral junction measuring about 8.70 cm × 3.45 cm × 8.60 cm in the craniocaudal, transverse, and anteroposterior dimensions. It had a mass effect on the lower ribs, which caused a chest wall deformity. There was also pseudo-articulation with the third rib. There was extension to the right hemithorax primarily at the level of the upper and middle lobes associated with adjacent middle lobe atelectatic changes. The intrathoracic extension of the exostosis was close to the right pulmonary vasculature. An intrathoracic extension portion measured 0.5 cm × 3.3 cm × 3.7 cm. Scattered exostoses were noted along the third, seventh, and eighth right ribs and the seventh left rib. Three-dimensional reconstruction was created to improve the understanding of the lesion and better guide the surgical operation (Figure 3).

FINAL DIAGNOSIS

Multiple bony lesions consistent with HME with a dominant large right chest wall lesion that caused significant pain and disability.

TREATMENT

Due to the growth of the chest wall lesion and the significant discomfort that the patient experienced, surgical resection of the main chest lesion with chest wall reconstruction using SurgiMend® biologic mesh (Integra LifeSciences Corp., Plainsboro, NJ, United States) was planned. This mesh is composed of acellular dermal matrix derived from fetal and neonatal bovine dermis. This mesh is durable, malleable, and capable of

allowing native tissue integration at the defect site. After discussion with the plastic surgeons, a latissimus dorsi muscle flap was planned to cover the mesh, if needed.

As shown in Figure 4A, the incision was marked over the center of the lesion. The plastic surgeon was present to ensure that the adjacent muscles and their neurovascular bundles were protected. The bony lesion was completely resected with the central portion of the third and fourth ribs (Figure 4B). The intrathoracic part of the lesion was not attached to the lung or mediastinal structures. The resulting chest wall defect was approximately 12 cm × 9 cm (Figure 4C). SurgiMend® mesh was then used to cover the defect in an onlay fashion using 0 prolene interrupted U-shaped sutures involving the surrounding ribs and intercostal muscles (Figure 4D). The reconstruction was satisfactory, and there was adequate soft tissue coverage. Therefore, the latissimus dorsi muscle flap procedure was unnecessary. Two closed suction drains were placed in the chest and in the subcutaneous tissue (Figure 5A). The *ex vivo* size of the lesion was 10.5 cm × 9.0 cm × 5.5 cm (Figure 5B).

Postoperatively, the patient was admitted to the pediatric intensive care unit and was extubated the next day. The drains were removed on the 3rd postoperative day, and the patient was sent home on the 6th postoperative day. Histopathological assessment of the resected specimen was consistent with osteochondroma without elements of malignancy.

OUTCOME AND FOLLOW-UP

The patient was followed up monthly in the pediatric surgery clinic. He was very cheerful and happy. He has had no complications, and the chest wall reconstruction remained satisfactory at the 1-year follow-up (Figure 6). No recurrence nor chest wall deformities were noted.

DISCUSSION

HME is a complex musculoskeletal disorder characterized by multiple osteochondromas primarily affecting long bones, ribs, and vertebrae. It is a rare

autosomal dominant disorder with an estimated incidence of 1:50000[1,2]. Over 80% of HME cases are associated with loss-of-function mutations in the *EXT1* or *EXT2* genes. Although exostoses are initially asymptomatic, they can cause significant health problems including pain, limb discrepancy, short stature, nerve or vessel impingement, skeletal deformities, and a restricted range of motion[1,2,5]. In approximately 2% of patients, osteochondromas can undergo malignant transformation to form chondrosarcomas or osteosarcomas[24]. The disease has no medical treatment. However, surgery is recommended for symptomatic exostosis or when malignant transformation is suspected[3,4]. Symptoms that may warrant surgical resection include significant pain or the occurrence of the aforementioned complications[12,25,26]. Several complications of chest wall osteochondromas have been reported in the literature. Some are life-threatening, including erosion into the adjacent structures causing hemothorax, pneumothorax, pericardial effusion, or diaphragmatic injury[6-11,14,26-30]. Rapid growth of the lesion may indicate malignant transformation[3,4,14].

The patient in this case report had significant chest pain due to the mass increasing in size. His pain caused significant problems with attending school and negatively affected his sleeping habits. Additionally, the rapid increase in the size of the dominant chest lesion has a risk factor for malignant transformation. The child and his parents were very interested in surgical treatment, even after discussion of the potential challenges to reconstruct the chest wall and the possible complications.

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Surgical resectability of chest wall lesions is determined by several factors including the number of ribs involved, the degree of intrathoracic extension, the involvement of the spinal canal, the reconstructability of the chest wall, and the possible functional outcomes after the resection. Cross-sectional imaging should be carefully studied during surgical planning to accurately define the extent of the tumor, plan the resection technique, and choose the appropriate reconstruction strategy. Three-dimensional reconstruction of the cross-sectional images provided a useful preoperative tool to study the surgical field.

In the operating room, the patient should be positioned so that the area to be resected is easily accessible. Access to the latissimus dorsi and pectoralis major muscles is important in cases where reconstruction with the muscle flap is required. In this case, the incision line was marked by the plastic surgeon to ensure that the neurovascular bundle of the major chest wall muscles was not compromised.

After the resection was completed, chest wall reconstruction was composed of two parts: Skeletal reconstruction and soft tissue coverage. Typically, resections involving less than four ribs do not require skeletal reconstruction. However, soft tissue reconstruction is still required in these situations. In contrast, larger defects (involving more than four ribs and/or defect size of > 5 cm) will require skeletal reconstruction with biologic or alloplastic material. The ideal reconstruction should perform the following functions: Restore chest wall rigidity; prevent lung herniation; avoid chest wall contraction; prevent trapping the scapula; protect underlying thoracic and mediastinal structures; and provide acceptable cosmetic results[15,17,20,26,31].

A wide variety of materials are now available for prosthetic chest wall reconstruction, including biologic, alloplastic, and synthetic materials. Table 1 provides a summary of the literature regarding chest wall reconstruction in children. The ideal prosthetic material for chest wall reconstruction should be rigid enough to prevent paradoxical chest wall movement, malleable to allow for contouring, able to grow with or accommodate the growth of the pediatric patient, biologically inert, stimulatory for tissue in-growth, radiolucent, resistant to infection, and relatively inexpensive. Several materials have been used successfully to reconstruct the chest walls of pediatric patients; however, no material has been proven to be superior in chest wall reconstruction in pediatric patients[20,32]. The materials include absorbable polyglactin, non-absorbable polypropylene, and polytetrafluoroethylene. Alternatively, biological meshes derived from cadaveric human dermis, porcine intestinal submucosa, porcine dermis, bovine pericardium, and bovine dermis (SurgiMend®; Integra LifeSciences Corp., Plainsboro, NJ, United States) have been used with acceptable results[14,16,21,22,26,32-35]. For robust reconstruction of very large defects, metallic rib substitutes have been

used in pediatric patients[19,36,37]. There have been no comparative studies of the different prosthetic materials, which should be a focus for future studies.

In this patient, SurgiMend®, which is composed of acellular dermal matrix derived from neonatal and fetal bovine dermis, was utilized for chest wall reconstruction. It is known to be durable as well as to conform to the shape of the chest wall. No mesh-related complications were observed within the 1-year follow-up period. The soft tissue coverage in this patient was acceptable without requiring the mobilization of muscle flaps. Based on our experience, we think that acellular dermal matrix mesh is an appropriate prosthesis to be used for chest wall reconstruction in children.

CONCLUSION

Exostosis, whether sporadic or caused by HME, may require surgical resection if it causes significant symptoms or disability. Moreover, chest wall lesions can be particularly more challenging in pediatric patients. Surgeons must carefully plan the most appropriate chest wall reconstruction strategy and choose from a variety of prosthetic materials. To assess late complications, such as prosthesis complications or acquired chest wall deformities, long-term follow-up is essential, particularly in pediatric patients.

Figure Legends

Figure 1 Image of the patient. A large irregular right chest wall mass was observed. Several smaller lesions were obvious in the lower part of the chest bilaterally.

Figure 2 Chest X-ray of the child showing a large opacity over the right side of the chest.

Figure 3 Computed tomography of the patient's chest with three-dimensional reconstruction showed a large irregular bony chest wall lesion with an intrathoracic extension.

igure 4 Intraoperative images of the resection and reconstruction. A: Transverse upper thoracic incision over the lesion; B: The lesion exposed; C: The chest wall defect after the lesion was completely resected; D: Biological mesh was placed to cover the defect.

Figure 5 Postoperative images. A: Two closed suction drains were placed after chest closure; B: The lesion after resection measured 10.5 cm × 9.0 cm × 5.5 cm.

Figure 6 The patient's chest wall 1 year after surgery with acceptable cosmesis.

Table 1 Summary of reported prosthetic material used for chest wall reconstruction in children

Ref.	Year of publication	Number of patients	Ages	Diagnoses	Prosthetic material used for reconstruction	Outcomes
Grosfeld *et al*[38]	1988	15	8 mo-11 yr	ES, RMS, NBM, CS, MS, Wilms' tumor	Marlex mesh, Gore-Tex® patch, Prolene mesh	Acceptable
Dang *et al*[39]	1999	4	10 wk-23 yr	ES, PNET, RMS, Askin's tumor	Marlex mesh, Gore-Tex® patch, Dexon mesh	Acceptable
Tuggle *et al*[40]	2004	4	6 mo–19 yr	ES, PNET, spindle cell tumor	Bioabsorbable copolymer plates (Lactosorb)	Acceptable
Soyer *et al*[41]	2006	9	11 mo-14 yr	ES, RMS, OC, NBM	Gore-Tex® patch, Neuro-patch, Dura	Acceptable (1 minor wound infection after Dura reconstruction)
Stephenson *et al*[19]	2011	6	5- 19 yr	ES, OS, Desmoid tumor, congenital thoracic insufficiency	Titanium constructs	Acceptable

				(Poland syndrome)		
Jackson *et al*[42]	2011	2	3 yr, 4 yr	PPB, infiltrative fibrous hamartoma of infancy	Gore-Tex® patch, STRATOS™ titanium bar	Acceptable
Dingemann *et al*[32]	2012	8	4-19 yr	ES, OS, PNET, RMS	STRATOS™ titanium bar, Gore-Tex® patch, Vicryl® patch, Tutopatch®	Acceptable
Lin *et al*[21]	2012	5	9-21 yr	ES, PNET	Biologic mesh (Permacol)	Acceptable
Makarawo *et al*[43]	2015	1	13 yr	ES	Polylactide bioabsorbable Struts (BioBridge®)	Acceptable
Lopez *et al*[18]	2017	22	4-18 yr	ES, RMS, CS, OS, SS	Gore-Tex® patch, Marlex mesh, Dexon mesh, SurgiMend® mesh, Vicryl® mesh	2 wound infections with exposed prosthesis required additional surgeries
Miyake *et al*[44]	2017	8	8-12 yr	ES	Bioabsorbable copolymer	Acceptable

			plates (Lactosorb)

ES: Ewing sarcoma; RMS: Rhabdomyosarcoma; NBM: Neuroblastoma; OC: Osteochondroma; CS: Chondrosarcoma; SS: Synovial sarcoma; MS: Mesenchymal sarcoma; PNET: Primitive neuroectodermal tumor; PPB: Pleuropulmonary blastoma.

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