Case Report

A Case of Giant Subcutaneous Bronchogenic Cyst

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Received: 19 May 2020; **Accepted:** 01 June 2020; **Published:** 03 July 2020

Abstract

Bronchogenic cysts are congenital anomalies that are typically found in the mediastinum or within the lung. Subcutaneous and cutaneous lesions are rare and most likely represent ectopic or displaced mesenchyme during early development. A 59-year-old male was referred by a respiratory surgeon for a giant tumor in the pre-sternal region. The tumor was elastic soft, round and mobile; however, the base was fixed to the sternal area. A CT scan showed round and cystic lesion of 14.7 × 12 × 8.0 cm in size and localized on the sternum. We began the tumor resection while draining fluid from the cyst slowly to maintain adequate tension on the surface on the cyst. We carefully dissected the surrounding tissue from the cyst to avoid rupture, and excised the cyst completely. There was no connection between the cyst and thorax. Unclear yellowish and seromucous fluid totaling 70 ml was drained. We closed the surgical wound inserting a drain tube after irrigation. Histopathological examinations revealed benign ciliated columnar epithelium with goblet cells located in the dermis and hypodermis, and partly lined by keratinizing stratified squamous epithelium. Glandular acini and smooth muscle exist within the cyst. The diagnosis was subcutaneous bronchogenic cyst. In this report, we describe an operation to treat a giant subcutaneous bronchogenic cyst 15 × 12 cm in size.

Keywords: Bronchogenic tissue; Presternal cyst; Heterotropic lung tissue

Abbreviations: CT- Computed tomography; H&E- Hematoxylin and Eosin

1. Introduction

Bronchogenic cysts are congenital anomalies that are typically found in the mediastinum or within the lungs [1]. Subcutaneous and Cutaneous lesions are rare and most likely represent ectopic or displaced mesenchyme during early development [2]. Seybold WD and Clagett OT first described the cutaneous bronchogenic cyst in the pre-sternal area in 1945 [3]. Fraga S, Helwig EB, and Rosen SH published a review of 30 cases of bronchogenic cysts in the skin and subcutaneous tissue in 1971 [4]. These lesions are mostly located in the suprasternal notch, pre-sternal area, shoulder and neck. The cysts are more common in male patients (83%). Approximately two thirds are present at birth. Kobayashi M, Soude E, Takahashi E, et al. published a review of 49 cases of subcutaneous bronchogenic cyst [5]. The biggest size was a 10 × 8 cm cyst on the suprasternal notch of a 58-year-old male patient. We removed a huge subcutaneous bronchogenic cyst 15 × 12 cm in size in the pre-sternal region as described in this report.

2. Case Report

A 59-year-old male was referred by a respiratory surgeon for a huge tumor in the pre-sternal region (Figure 1). The tumor was elastic soft, round and mobile; however, the base was fixed on the sternal area. He had undergone pulmonary cancer operation six months before. The right lower lobectomy of the lung and lymph node dissection were performed under thoracoscopic surgery due to adenocarcinoma (pT1cN0). The tumor on the pre-sternal region which had been present since birth gradually became bigger in size with age. The patient did not complain of any pain around the tumor and of any swallowing disorder.

A CT scan showed round and cystic lesion 14.7 × 12 × 8.0 cm in size and localized on the sternum. The image diagnosed a mature teratoma. No abnormalities were observed in the lungs, thymus, thyroid and other organs. He desired to undergo the surgical resection. We, plastic surgeons, decided and marked double lazy S incision lines to avoid keloid (especially sternal region) and to remove the thin brownish pigmented skin on the center of the tumor, and added a skin incision. The intravenous catheter was fixed into the cystic lesion for drainage of the fluid contents and connected to a tube with a three-way stopcock which was attached to a syringe. We began the tumor resection while draining the fluid from the cyst slowly because it is important to keep adequate tension on the surface of the cyst. While pulling up the cyst and maintaining the tension, we carefully dissected the surrounding tissue from the cyst to avoid rupture of the capsule, and excised the cyst completely (Figure 2). There was no connection between the cyst and thorax. Unclear yellowish seromucous fluid totaling 70 ml was drained. We closed the surgical wound inserting a drain tube after irrigation.

Histopathological examination of the cyst with H&E staining revealed benign ciliated columnar epithelium with goblet cells, which was located in the dermis and hypodermis, and partly lined by keratinizing stratified squamous epithelium. Glandular acini and smooth muscle were found within the cyst (Figure 3). The diagnosis was a

subcutaneous bronchogenic cyst. The patient had no complications after operation. A CT scan showed no recurrence of the tumor 6 months after operation (Figure 4).

Figure 1: Giant tumor on the pre-sternal region of a 59-year-old male. The tumor was elastic soft, round and mobile; however, the base was fixed on the sternal area.

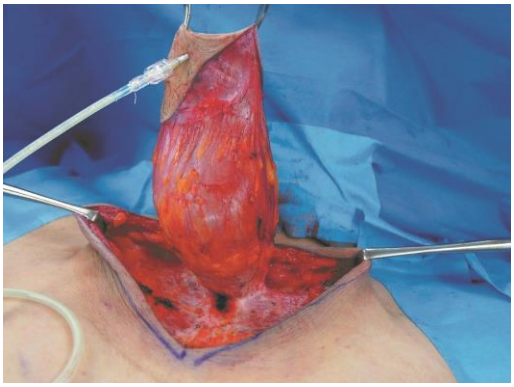

Figure 2: Gant cyst on the pre-sternal region. There was no connection between the cyst and thorax. An intravenous catheter was fixed into the cystic lesion for drainage of the fluid contents. A tube was connected between the catheter and a three-way cock, which was attached a syringe.

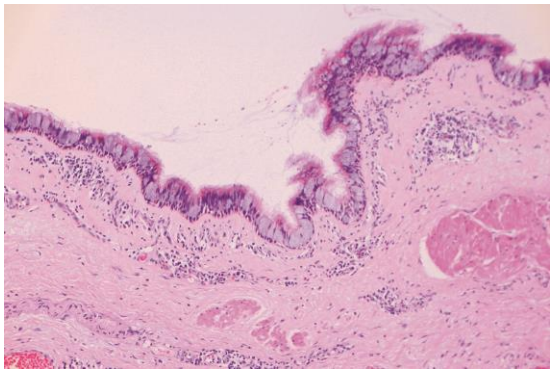

Figure 3: H.E stain of the cyst showed benign ciliated columnar epithelium with goblet cells located in the dermis and hypodermis and partly lined by keratinizing stratified squamous epithelium.

Figure 4: Condition 6 months after operation.

3. Discussion

Bronchogenic cysts are rare congenital anomalies that are usually located in the mediastinum or lung parenchyma. Bronchogenic cysts in the subcutaneous region are very rare and thought to result from abnormal development of the tracheobronchial system [6]. An abnormal budding of the tracheobronchial system between the 22nd and 33rd days of gestation and persistence of such a bud may give rise to bronchogenic cyst. Abnormal migration of a bud may occur during the course of development and rest in different intrathoracic or extra-thoracic locations[6]. In another theory, the bronchogenic cyst, which already exists, is accepted to be left out of the thorax after sternal closure and migrates to the cutaneous region [7]. Fraga S, Helwig EB, and Rosen SH published a review of 30 cases of bronchogenic cysts in the skin and subcutaneous tissue in 1971. The lesions varied in diameter from 0.3 to 6 cm [7]. Kobayashi M et al. published a review of 49 cases of intracutaneous bronchogenic cysts in 2008 [5]. She reported that the biggest cyst on the suprasternal notch measured size 10 × 8 cm. Inoue S et al. published a review of 107

cases in 2019 [8]. The lesions varied from 0.5 to 5.5 cm. A subcutaneous cyst measuring 15 cm is huge and has not ever been reported previously.

However, the cyst existed in the presternal region. Therefore, long follow-up must be continued because the postoperative scar might change to keloid or hypertrophic scar formation. The distinct histologic features are similar to those of the bronchogenic cysts in intrathoracic locations, including the presence of ciliated pseudostratified columnar epithelium, usually of smooth muscle and seromucous glands, and of cartilage in a few cases [4]. Malignant transformation of a bronchogenic cyst has been rare [9]. A case of malignant melanoma arising from a cutaneous bronchogenic cyst of the left scapular area was reported. Suen HC et al. reported adenocarcinoma arising from a bronchogenic cyst in an 8-year-old girl and the case of a malignant pleural mesothelioma, accompanied by a bronchogenic cyst [10]. The differential diagnosis for the cyst includes branchial cleft cyst, thyroglossal duct cyst, cutaneous ciliated cyst, and matured cystic teratoma. They can be differentiated from the site of the development as well as by each unique histopathologic feature [9].

4. Conclusion

We have described a case of giant subcutaneous bronchogenic cyst.

Conflicts of Interest

There are no conflicts of interest to declare.

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Citation: Takako Nakao, Misato Katayama, Hiromi Kino, Nobuharu Hanaoka, Koichi Ueda. A Case of Giant Subcutaneous Bronchogenic Cyst. Archives of Clinical and Medical Case Reports 4 (2020): 590-595.