



Case report

Subcutaneous bronchogenic cyst of the chest wall: A case report with brief literature review

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ABSTRACT

Introduction: Bronchogenic cysts are congenital lesions found in the mediastinum, particularly the posterior-superior area. The current study aims to report a rare case of a subcutaneous bronchogenic cyst in the chest wall. **Case report:** A 41-year-old patient presented with a swelling of the chest wall. The mass had been present since birth. On examination, there was a large soft, round mass over the sternum subcutaneously. It was a fixed, non-flatulence, non-pulsatile, and non-tender mass.

Discussion: Usually, the condition develops between the fifth and sixteenth weeks of gestation, when the primordial intestine separates into two parts: dorsal, which gives rise to the esophagus, and ventral, which gives rise to the pulmonary bud and tracheobronchial tree. As a result, the cyst is an ectopic lung bud that may or may not be connected to the tracheobronchial tree but lacks mesenchymal tissue.

Conclusion: Although chest wall bronchogenic cysts are uncommon, they should be considered in the differential diagnosis of cystic and soft tissue lesions in adults with chest wall swelling.

1. Introduction

Bronchogenic cysts are congenital lesions caused by defective ventral foregut budding during tracheal and major bronchial structural development [1]. Bronchogenic cysts are rare, with a frequency of 1 in 42–68,000 people. When they occur, the majority are found in the mediastinum, particularly the posterosuperior area [2]. Intrapulmonary cysts account for approximately 15–20% of all bronchogenic cysts and are typically found in the lower lobes. They have been observed not just in infants and children but in adults as well later in life [3]. They can form in ectopic positions anywhere along the foregut developmental pathway [4]. On rare occasions, they have been observed in atypical sites such as the scapula, paravertebral, cervical, retroperitoneal, pericardial, omental, and perianal regions [5]. A chest wall bronchogenic cyst is an extremely rare condition, with just a few occurrences previously recorded in the literature [4–6].

The current study aims to describe a case of a subcutaneous bronchogenic cyst. The report is organized in line with SCARE 2020 criteria

and contains a brief review of the literature [7].

2. Case report

2.1. Patient's information

A 41-year-old male patient presented with a swelling of the chest wall. The mass had been present since birth, but it grew in size later in life till it burst 15 years ago. Three months after it burst, it began to gradually grow in size till the current presentation to the clinic. Dyspnea and chest discomfort are common symptoms. He has a history of asthma and underwent PCI in 2019. He is now taking aspirin, Plavix, and an inhalational short-acting beta-blocker as needed. He is not a smoker.

2.2. Clinical examination

On examination, there was a large (7 cm * 7 cm), soft, round mass over the sternum subcutaneously. It was a slightly mobile, non-

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flatulence, non-pulsatile, and non-tender mass. There was no discoloration of the overlying skin, and the temperature was normal.

2.3. Diagnosis

The provisional diagnosis was lipoma. No investigation was performed, the patient insisted on the removal of the mass.

2.4. Therapeutic intervention

Under local anesthesia, a transverse incision exposed a cystic lesion containing milky fluid and adhering to the chest wall. Complete excision was performed, a corrugate drain was placed, and the wound was closed in layers. Histopathological examination revealed fibrofatty tissue transversed by cystic space and lined by cuboidal to pseudostratified ciliated columnar epithelium (respiratory type) resting on a fibrous stroma. The overall picture goes with a subcutaneous cyst of the chest wall.

2.5. Follow up

The patient had a non-eventful postoperative period. After one week, the stitches and drain were removed without complications.

3. Discussion

Congenital cystic lung lesions, which include congenital cystic adenomatoid malformation, pulmonary sequestration, congenital lobar emphysema (CLE), and bronchogenic cysts, are an uncommon but clinically significant set of anomalies [8]. Bronchogenic cysts are non-malignant congenital anomalies of the primordial ventral foregut. The cyst wall contains structural characteristics of the airway such as cartilage, smooth muscle, mucous glands, and respiratory epithelium [9,10]. They are most commonly found intra-thoracic and are classified as mediastinal or parenchymal, depending on where they are found. However, they can also be found intra-abdominal or, in rare cases, cervical [11]. Up to 86% are mediastinal (middle and posterior mediastinum), and of these, some may be adjacent to the distal third of the trachea or close to the main bronchus; thus, they can be subdivided into peri-carinal, paratracheal, para-esophageal, and retro-cardiac, with the majority being in the right [12]. They've been found attached to the sternum, pericardium, skin, and even the diaphragm [13].

The precise method through which bronchogenic cysts become subcutaneous is unknown. Proposed mechanisms include the aberrant tracheobronchial bud being separated from the respiratory tract by growing skeletal structures and the bronchogenic cyst migrating into the subcutaneous tissues [14]. They are typically unilocular and contain clear fluid or, less frequently, hemorrhagic secretions or air [15].

Bronchogenic cysts present clinically and radiologically in a variety of ways, ranging from incidental radiologic findings without symptoms to giant mass-like structures accompanied by severe symptoms [16,17]. The variability of presentation depends on their size, location, and compression or invasion of adjacent tissues [5]. According to one study, 71% of children with mediastinal cysts were symptomatic [18]. A cough, difficulty breathing or swallowing, hemoptysis, and infection are symptoms of enlarged cysts. Symptoms of cutaneous bronchogenic cysts include pain, a growing mass, a draining sinus, and, in rare cases, cellulitis and abscess [5]. Lee et al. reported that more than half of their cases were symptomatic, with cough being the most common symptom [17]. Almost always, subcutaneous bronchogenic cysts are asymptomatic. The only symptoms reported in the cutaneous bronchogenic cysts are nasal puffiness and discharge [19]. Shilova et al. reported a case of a pre-sternal bronchogenic cyst with tender, erythematous swelling [14]. Sirvani et al. reported a 6-year-old male child who presented with a pre-sternal discharge sinus [20].

Although no modality has proven particularly specific for the

diagnosis of bronchogenic cysts, pre-operative imaging such as ultrasonography, CT, and MRI has been employed to better describe these masses. Plain radiographs have minimal use. A unilocular fluid-filled cystic tumor is often seen on ultrasonography [21]. CT and MRI provide excellent details about the size of the cyst and its connection to adjacent tissues, which is very useful in pre-operative planning [22]. CT usually shows an encapsulated mass without contrast enhancement and various levels of fluid attenuation, but MRI shows enhancement on T2-weighted images owing to mucinous and proteinaceous debris [23]. After a comprehensive clinical assessment, no imaging was deemed necessary in the current case. A histopathological examination confirms the definitive diagnosis, which shows a cyst with ciliated pseudostratified columnar epithelium, indicating a respiratory origin. The histopathology distinguishes it from other types of cystic masses, such as epidermoid cysts, teratomas, lymphangiomas, and dermoid cysts [24]. The same findings were observed in the current case.

The differential diagnosis for bronchogenic cysts is established by their location in the body. A cystic neck mass might be an abscess, a thyroglossal duct cyst, or a bronchogenic cyst [25]. A cystic chest wall mass can be diagnosed as a dermoid cyst or teratoma, and a mediastinal cyst can be TB, foregut cyst, pericardial cyst, congenital cystic adenoid malformation, pulmonary sequestration, large B cell lymphoma, or enterogenic cyst [6].

All bronchogenic cysts should be excised owing to the danger of the consequences such as enlargement, compression of adjacent tissues, infection, and cancer (bronchioalveolar carcinoma, adenocarcinoma, squamous cell carcinoma, and melanoma) [18,26]. Patients with bronchogenic cysts have a 0.7% lifetime risk of developing cancer [18]. This supports the choice of early excision as soon as possible. The current case underwent surgical excision as recommended.

In conclusion, although chest wall bronchogenic cysts are uncommon, they should be considered in the differential diagnosis of cystic and soft tissue lesions in adults with chest wall swelling. For a definitive diagnosis and avoiding possible consequences, surgical resection is suggested.

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None is found.

Ethical approval

Approval is not necessary for case report (till 3 cases in single report) in our locality.

The family gave consent for the publication of the report.

Consent

Written informed consent was obtained from the patient's family 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.

Author contribution

Abdulwahid M. Salh: major contribution of the idea, literature review, final approval of the manuscript.

Sangar Abubakir A. Mirawdali: Surgeon performing the operation, final approval of the manuscript.

Fahmi H. Kakamad, Marwan N. Hassan: Writing the manuscript, literature review, final approval of the manuscript.

Hiwa O. Baba, Fattah H. Fattah, Karzan M Salih, Shvan H Mohammed: literature review, final approval of the manuscript.

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None to be declared.

References

- [1] T.M. Monaghan, J.D. Thomas, T. Hussain, A bronchogenic cyst causing chest pain and dysphagia, *QJM* 104 (6) (2011) 539–541.
- [2] E. Ustundag, M. Iseri, G. Keskin, B. Yayla, B. Muezzinoglu, Cervical bronchogenic cysts in head and neck region: review of the literature, *J. Laryngol. Otol.* 119 (6) (2005) 419–423.
- [3] A. Kosar, C. Tezel, A. Orki, H. Kiral, B. Arman, Bronchogenic cysts of the lung: report of 29 cases, *Heart Lung Circ.* 18 (3) (2009) 214–218.
- [4] D.H. Lee, T.M. Yoon, J.K. Lee, S.C. Lim, Bronchogenic cyst in the head and neck region, *J. Craniofac. Surg.* 28 (4) (2017) e303–5.
- [5] A. Ayub, A.M. Abid, S. Tran, K. Bowen-Jallow, Subcutaneous bronchogenic cyst of the chest wall, *J. Pediatr Surg Case Rep* 52 (2020), 101337.
- [6] J.E. Cohn, K. Rethy, R. Prasad, J. Mae Pascasio, K. Annunzio, S. Zwillenberg, Pediatric bronchogenic cysts: a case series of six patients highlighting diagnosis and management, *J. Investig. Surg.* 33 (6) (2020) 568–573.
- [7] R.A. Agha, T. Franchi, C. Sohrabi, G. Mathew, for the SCARE Group, The SCARE 2020 guideline: updating consensus Surgical CAse REport (SCARE) guidelines, *Int. J. Surg.* 84 (2020) 226–230.
- [8] L.T. Rios, E. Araujo Júnior, L.M. Nardoza, A.F. Moron, M.D. Martins, Prenatal diagnosis and postnatal findings of bronchogenic cyst, *Case Rep. Pulmonol.* 2013 (2013), 483864.
- [9] P. Cuyppers, P. De Leyn, L. Cappelle, L. Verougstraete, M. Demedts, G. Deneffe, Bronchogenic cysts: a review of 20 cases, *Eur. J. Cardiothorac. Surg.* 10 (6) (1996) 393–396.
- [10] A.N. Kumar, Perinatal management of common neonatal thoracic lesions, *Indian J. Pediatr.* 75 (9) (2008) 931–937.
- [11] J.K. Mattingly, J.M. Arganbright, M.A. Lovell, K.H. Chan, Cervical bronchogenic cysts: case report and review of the literature, *Am. J. Otolaryngol.* 35 (5) (2014) 655–657.
- [12] C.A. Quezada-Salazar, M. Navarrete-Arellano, Bronchogenic cyst, prenatal diagnosis, *Bol. Med. Hosp. Infant. Mex.* 62 (3) (2005) 202–206.
- [13] R.J. Wirbel, U. Uhlig, E.M. Kiffner, K. Berger, Bronchogenic cyst as a rare differential diagnosis of retroperitoneal tumor, *Chirurg* 64 (12) (1993) 1056–1059.
- [14] M. Shilova, M.C. Wong, J. Davies, R. Kimble, A bronchogenic cyst presenting as a presternal abscess, *J. Pediatr Surg Case Rep* 63 (2020), 101653.
- [15] A. Sarper, A. Ayten, I. Golbasi, A. Demircan, E. Isin, Bronchogenic cyst, *Tex. Heart Inst. J.* 30 (2) (2003) 105.
- [16] D.H. Lee, C.K. Park, D.Y. Kum, J.B. Kim, I. Hwang, Clinical characteristics and management of intrathoracic bronchogenic cysts: a single center experience, *Korean J. Thorac. Cardiovasc. Surg.* 44 (4) (2011) 279.
- [17] C.A. Efthymiou, E.M. Kefaloyannis, J.A. Thorpe, Massive bronchogenic cyst mimicking ischaemic chest pain, *Eur. J. Cardiothorac. Surg.* 34 (6) (2008) 1260–1261.
- [18] J.H. Jiang, S.L. Yen, S.Y. Lee, J.H. Chuang, Differences in the distribution and presentation of bronchogenic cysts between adults and children, *J. Pediatr. Surg.* 50 (3) (2015) 399–401.
- [19] V. Manchanda, A. Mohta, N. Khurana, S. Das, Subcutaneous bronchogenic cyst, *J. Cutan Aesthet Surg* 3 (3) (2010) 181.
- [20] N. Srivani, S. Srujana, R. Prasad, L. Krishna, O.S. Kumar, Presternal cutaneous bronchogenic cyst: a rare case, *Int. Surg. J.* 1 (3) (2016) 148–151.
- [21] N. Teissier, M. Elmaleh-Bergès, L. Ferkdadjji, M. François, T. Van Den Abbeele, Cervical bronchogenic cysts: usual and unusual clinical presentations, *Arch. Otolaryngol. Head Neck Surg.* 134 (11) (2008) 1165–1169.
- [22] R.P. Mehta, W.C. Faquin, M.J. Cunningham, Cervical bronchogenic cysts: a consideration in the differential diagnosis of pediatric cervical cystic masses, *Int. J. Pediatr. Otorhinolaryngol.* 68 (5) (2004) 563–568.
- [23] H.P. McAdams, W.M. Kirejczyk, M.L. Rosado-de-Christenson, S. Matsumoto, Bronchogenic cyst: imaging features with clinical and histopathologic correlation, *Radiology* 217 (2) (2000) 441–446.
- [24] Z.M. Mir, A. Wang, A. Winthrop, M. Kolar, Scapular bronchogenic cyst in a girl presenting as recurrent cellulitis: a case report and review of the literature, *Case Rep. Pediatr.* 2018 (2018).
- [25] W.C. Che, Q. Zang, Q. Zhu, T.C. Zhen, G.Z. Su, P. Liu, H.J. Ji, Lipoma-like bronchogenic cyst in the right chest sidewall: a case report and literature review, *Ann. Thorac. Cardiovasc. Surg.* 22 (6) (2016) 370–374.
- [26] M. De Perrot, J.C. Pache, A. Spiliopoulos, Carcinoma arising in congenital lung cysts, *Thorac. Cardiovasc. Surg.* 49 (03) (2001) 184–185.