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Original Research

Broncogenic Cyst in Atypical Locations: A Case Series

Ivonete Siviero, Ph.D, M.D.[1], Betina Carla Bertrand Simões, M.D.[1], André Lima da Cunha, Ms.C[1], Deborah Câmara Sciani, M.D.[1]

[1] Surgery, Federal University of Rio de Janeiro

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Context

Bronchogenic cysts are rare congenital malformations of the embryonic foregut, typically located in the mediastinum. This report presents three pediatric cases of subcutaneous bronchogenic cysts in atypical locations—the sternal manubrium, middle cervical region, and scapular area—contributing to the literature by highlighting the anatomical variability of these lesions and the challenges associated with preoperative etiological diagnosis.

Case Reports

The patients presented with subcutaneous masses exhibiting nonspecific clinical features, such as intermittent drainage, cutaneous appendages, and localized tenderness. Initial clinical impressions suggested alternative etiologies, including branchial remnants or dermoid/sebaceous cysts. In all three cases, definitive diagnosis was established through histopathological examination, which revealed pseudostratified ciliated columnar epithelium consistent with bronchogenic cysts. Surgical management involved complete excision of the lesions, with favorable postoperative outcomes and no recurrence.

Conclusion

Bronchogenic cysts may present in uncommon subcutaneous locations, complicating preoperative etiological diagnosis. Complete surgical excision is curative and prevents potential complications. These cases underscore the importance of including bronchogenic cysts in the differential diagnosis of pediatric cervical and subcutaneous masses, even when located in atypical regions.

INTRODUCTION

Bronchogenic cysts are rare benign congenital malformations of the embryonic foregut, defined by the presence of respiratory-type epithelium in their wall. They are believed to arise from epithelial cells that separate from the developing tracheobronchial tree.[1] Although they typically occur within or adjacent to the tracheobronchial tree, i.e. in the mediastinum or lung parenchyma, BC can be found in atypical sites.[1,2] Symptoms are related to the region where they manifest. Most of the time they are asymptomatic, but they can be complicated by infection, compression of neighboring structures or even malignant transformation.[1,2] We describe three patients with subcutaneous bronchogenic cysts (BC), located on the sternal manubrium, middle cervical region and scapular region, with special emphasis on the difficulties of preoperative etiological diagnosis and complications. They were treated by complete excision of the cysts and without recurrence.

CASE REPORTS

Case 1: a 3-year-old male patient with a history of an anterior cervical tumor seen for the first time five months previously. On examination, he had a tumor measuring 2 centimeters (cm) in length, with a cutaneous appendix, in the middle third of the neck, on the anterior border of the left sternocleidomastoid muscle, mobile, elastic in consistency, with no phlogistic signs or drainage orifice (Fig. 1.a). There were small ipsilateral lymph nodes. The suspected diagnosis was a cyst and a branchial remnant. During surgery, a cystic tumor was found at the edge of the sternocleidomastoid extending to the confluence of the carotid sinus, which was excised (Fig. 1.b). Histopathological examination revealed a cystic structure, one and a half centimeters long, pale brown with mucoid material inside and lined by ciliated pseudostratified columnar epithelium, compatible with a bronchogenic cyst BC). The patient evolved without any post-operative complications or recurrence in 10 years.

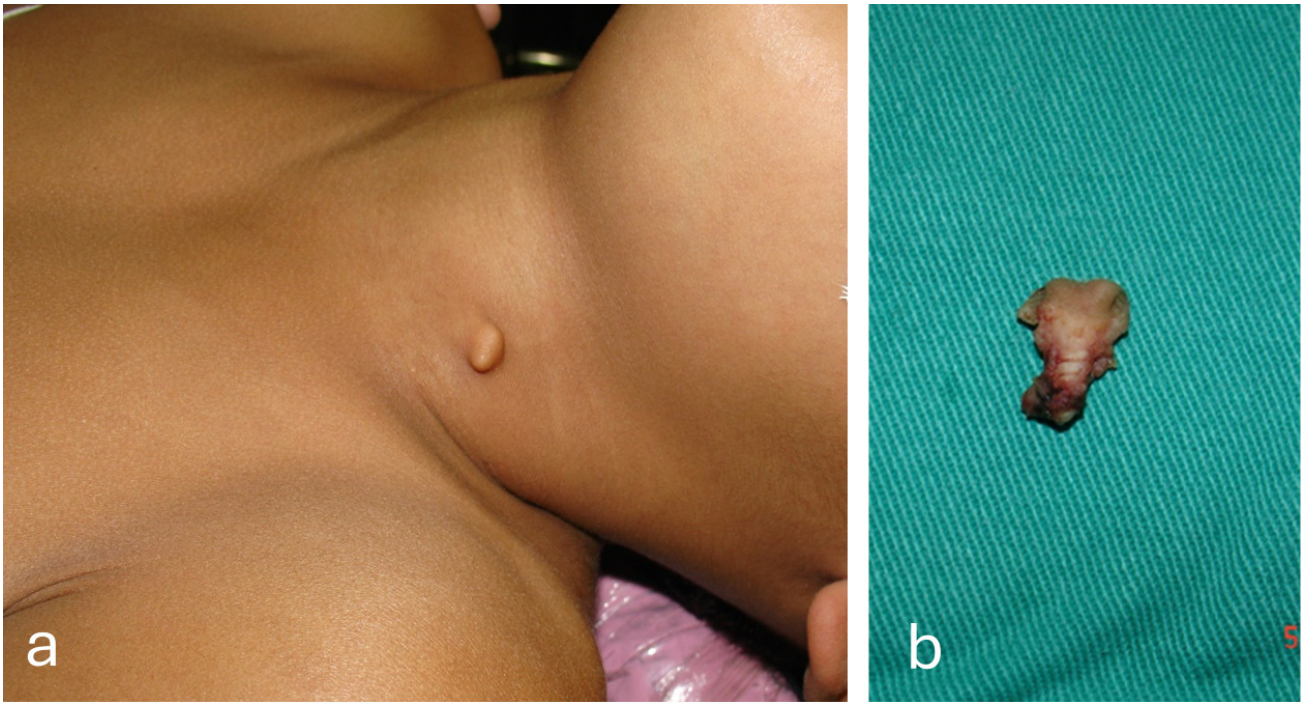

Figure 1. a) Cutaneous appendix on the anterior edge of the left sternocleidomastoid muscle. b) Resected surgical specimen.

Case 2: A seven-month-old male infant was seen due to a suprasternal sinus with sporadic discharge of hyaline secretion. On examination, he was in good general condition and presented with a cutaneous and subcutaneous, suprasternal sinus, one cm long with a drainage hole (Fig.2.a). The suspected diagnosis was a branchial cleft cyst. Surgery revealed the presence of a cystic tumor with a fistulous path to the sternal manubrium (Fig.2.b). Histopathological examination revealed a grayish-brown cystic structure with a rough surface lined with pseudostratified ciliated columnar epithelium compatible with BC. The fistulous tract was lined with stratified squamous epithelium. He evolved without complications or recurrence in 7 years.

Case 3: a 2-year-old male patient had a lesion in the right scapular region noticed 6 months previously and a history of sporadic spontaneous drainage. Ultrasound showed a cystic nodular image with regular borders, clear boundaries, with acoustic reinforcement by thick content, measuring 2.3 cm deep by 1.3 cm wide in the subcutaneous cellular tissue, suggesting the diagnosis of a dermoid or sebaceous cyst. In the right scapular region, there was an area of hyperemia with a small opening and purulent drainage. The surrounding skin was sensitive, but there was no fluctuation. He was given antibiotic therapy and underwent complete excision of the lesion and local skin in bloc. During surgery, it was noted that the cyst extended deeply, in close contact, but without continuity with the scapula. Histopathological examination revealed a BC, the wall of which was lined with pseudostratified ciliated columnar epithelium, supported by connective tissue. It contained smooth muscle, mucous glands and a lymphoid infiltrate. He has progressed without recurrence in three years of follow-up.

Table 1 summarizes the characteristics of the three cases.

DISCUSSION

Bronchogenic cyst has an incidence of 1 in 42,000 to 68,000 individuals.[3] A study carried out by Jyun-Hong Jiang et al showed that there are two peaks of distribution by age, one at 6 years and the other between 20-70 years.[1] They are thought to arise from an embryogenic failure of the anterior intestine to divide into the dorsal esophagus and ventral trachea. The primitive respiratory tissue can separate from the tracheobronchial tree or develop abnormally as a sprout, leading to the formation of bronchogenic cysts.[3,4] Due to their origin, most bronchogenic cysts are in the mediastinum. However, a BC with detached bronchogenic tissue can also be found in the chest wall, neck, diaphragm, heart, intra-abdominal and retroperitoneal areas.[1,4-8] It is not yet clear how these cysts reach these aberrant positions, such as the intergluteal fold.[9] After the thoracic cavity, the abdomen and subcutaneous tissue are the most common locations.[2]

BCs of the skin and subcutaneous cellular tissue are more frequently found in males, in a ratio of 4:1.[5] Most cases occur in early childhood, but there are reports at all ages.[1,5] Subcutaneous BCs are most often located in the neck, followed by the supra sternal notch, pre sternal region and scapular region.[1,5,9] The signs and symptoms that characterize these cysts are not specific, which makes their presumptive etiological diagnosis practically impossible.

Large cysts in the mediastinum can cause compression of the tracheobronchial tree, resulting in dyspnea or cough, or can be noticed on imaging tests. Cervical BCs are usually

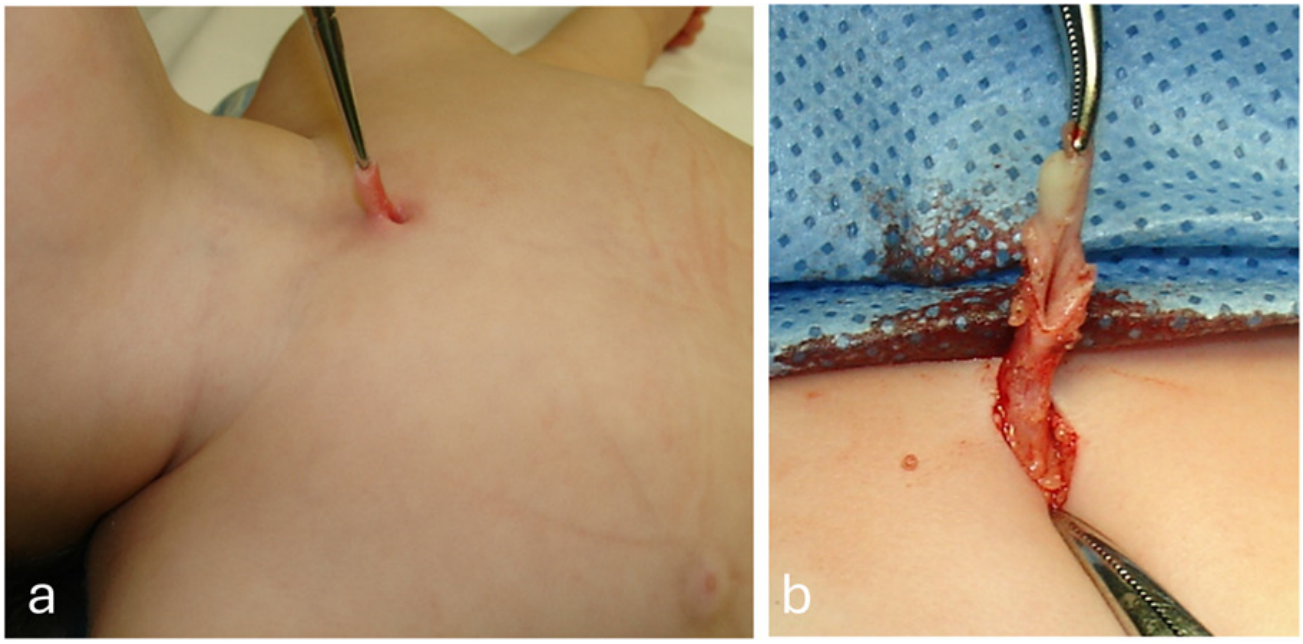

Figure 2. a) cutaneous appendix and sinus in the supra sternal region. b) photograph of the surgical specimen before it was completely resected.

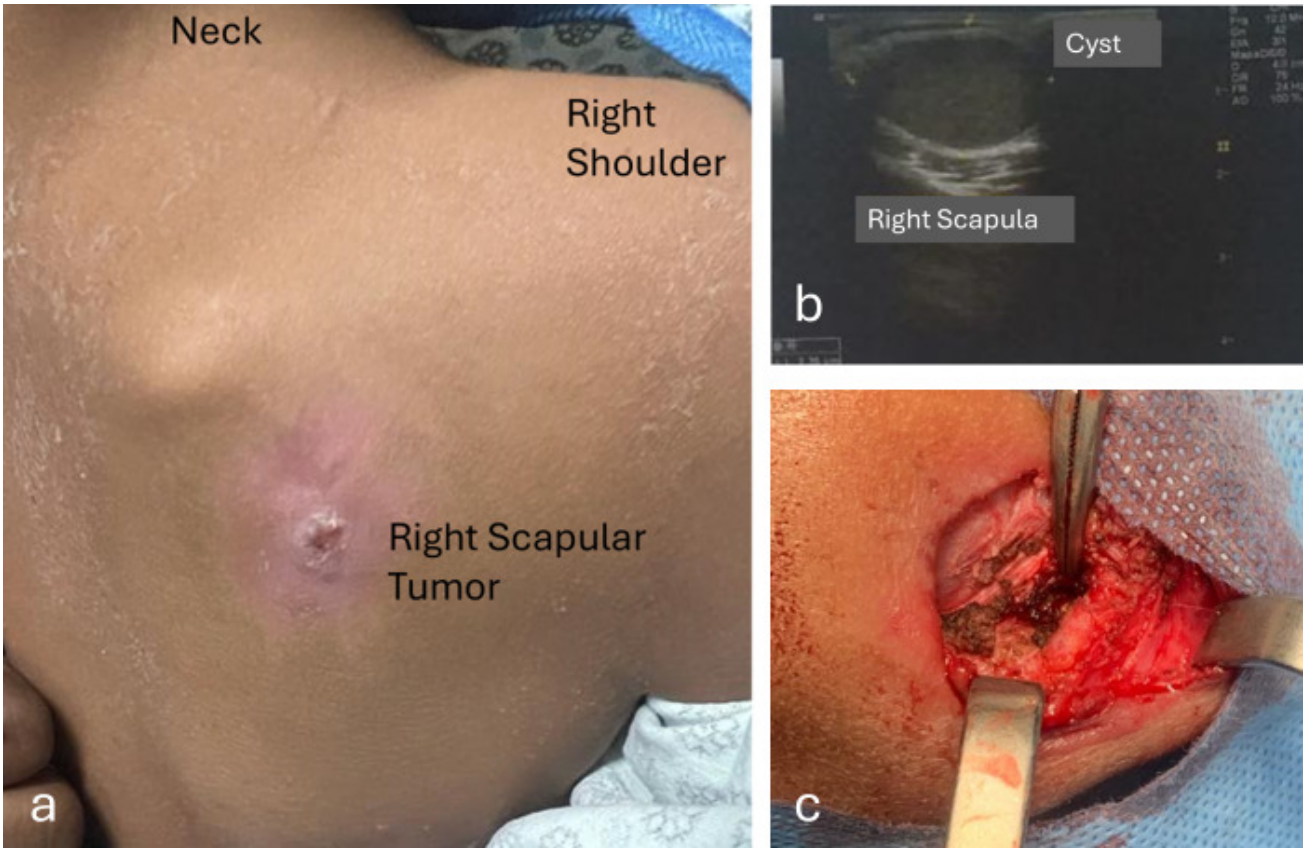

Figure 3. a) right scapular tumor with a history of previous spontaneous drainage; b) preoperative ultrasound showing a 23 x 16 mm cyst in the right scapular region; c) photograph of the surgical resection showing intimate contact with the scapula (Kelly clamp).

discovered during childhood and can manifest anywhere in the neck, including the midline and supra sternum.[10] Although they can be complicated by infection or compression of structures due to mass effect, they are usually asymptomatic and noticed as a cervical cystic tumor of varying size in childhood,[1,10] as in the two cases presented

Table 1. Clinical profile and evolution of the three patients

Case	Age	Sex	Location and Size	Preoperative Imaging	Surgical Approach	Histological Features	Follow-up (years)
1	3 years	Male	Neck, 2 cm in length	None	Complete excision	Pseudostratified ciliated columnar epithelium; absence of cartilage; presence of mucous glands	10
2	7 months	Male	Suprasternal region, 1 cm deep	None	Complete excision of fistulous tract	Pseudostratified ciliated columnar epithelium; fistulous tract lined by stratified squamous epithelium; absence of cartilage	7
3	2 years	Male	Scapular region, 2.3 cm deep by 1.3 cm wide	US: cystic nodular image	Complete block excision	Pseudostratified ciliated columnar epithelium; presence of mucous glands, smooth muscle, and lymphoid infiltrate; absence of cartilage	3

here. When located deeper in the cervical region or extending into the mediastinum, they can cause respiratory symptoms due to tracheal compression.[2,11] A superficial BC can become infected and establish a fistulous tract or even have a tract that connects it to the skin. More rarely, they present with an associated appendix, as in the two cervical cases reported in this study. There is a report of an subcutaneous BC present in a congenital cervical contracture in the midline of the neck.[12] Those in the scapular region, where it is extremely rare, can present as a slow-growing asymptomatic superficial tumor, with or without a cutaneous fistula, or as an acute inflammation and infection.[8] In one of the cases presented, the child had a local infection which complicated with cellulitis in the scapular region, making treatment difficult. This is not uncommon when it occurs in the scapular region.

As BC is rare in the cervical region, it is often confused with other more frequent congenital cysts in the neck. In the largest series of cervical cysts reported, the thyroglossal duct cyst was the most common, followed by lymphatic cysts, while the BC corresponds to only around 1% of cases.[13] The differential diagnosis also includes branchial cleft cysts, dermoid cysts, congenital cervical cleft, lipoma and teratoma.[1,10,14] If only cervical ciliated cysts are considered, the differential is reduced to a branchial cyst, thyroglossal duct cyst and thymic cyst.[5] Those in the scapular region are usually diagnosed as abscesses, dermoid cysts, lymphatic or vascular malformations or even lipomas.[8]

Currently, there are no specific imaging standards for BC. No imaging test can help lead specifically to a diagnosis, but they can help exclude other non-cystic pathologies or neoplasms. Ultrasound will show a cystic and anechoic image. Computed tomography and nuclear magnetic resonance provide excellent details on the extent of the cyst and its relationship with adjacent structures, which are useful in preoperative planning, especially when it is deeper.[10,11,14] In cases of subcutaneous BC, they are of little use and are usually not performed.

A definitive diagnosis can only be made by histopathology. Therefore, complete surgical excision is recommended, even for asymptomatic BC, to allow a definitive pathological diagnosis, as well as preventing complications and the potential risk of malignant transformation. BC has a thin wall and are filled with mucoid content. The wall has ciliated pseudostratified columnar epithelium covering the inner surface of the cyst since its origin is respiratory, which was confirmed in the three cases reported here. The cyst wall may also have fibroelastic tissue, mucous glands, smooth muscle and, more rarely, cartilage.[5,10,14]

Rarely, there is squamous metaplasia, ulceration of the mucosa, and various degrees of inflammation which can make it difficult or impossible to identify all the classic features,[14] especially in those cases with a history of previous infection.

In such cases, diagnosis relies on the careful evaluation of additional histological structures, such as mucous glands and smooth muscle, to support the identification of BCs, and on the exclusion of characteristic markers of other cyst types, namely, lymphoid tissue found in branchial cysts and cutaneous elements such as hair follicles, sebaceous glands, and adipose tissue typical of dermoid cysts.[5,14] Therefore, accurate histological correlation is essential to avoid misdiagnosis, particularly in atypical locations such as the subcutaneous tissue. Malignant transformation has been reported in less than 1% of cases in the adult population.[3,5,9,14,15]

Histochemical analysis is generally not required for the diagnosis of BCs and was not employed in these cases, as the diagnosis was established through conventional histopathological examination. This revealed classic features of BCs, including ciliated columnar epithelium, connective tissue, mucous glands, and smooth muscle, elements sufficient to distinguish them from branchial and dermoid cysts without the need for additional techniques.[3,5,14]

The treatment of BC consists of complete excision to prevent recurrences, paying special attention to the relationship with the structures of the neck, especially when located more deeply and including cartilage, if any, at the base.[2] The post-operative course of our cases was uneventful and there were no recurrences.

CONCLUSIONS

Although rare, bronchogenic cysts should be considered in the differential diagnosis of cystic tumors of the neck,

scapular region or any other region of the trunk. Imaging can be important for surgical planning, but the diagnosis can only be made by histopathological analysis. Treatment is complete excision.

ETHICAL CONSIDERATIONS

This study was approved by the Research Ethics Committee of the Martagão Gesteira Institute of Childcare and Pediatrics at the Federal University of Rio de Janeiro, under approval number 7.714.028 dated July 17, 2025. The patients provided informed consent for the publication of this case series.

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