Bronchogenic cysts are generally detected shortly after birth or in early childhood. These lesions are benign congenital developmental anomalies of the tracheobronchial buds from the primitive foregut, and the most common extrapulmonary location of this lesion is the mediastinum. Cutaneous bronchogenic cysts are rarely reported. The most common location is the suprasternal notch, followed by the presternal area, neck, and scapula. We report here on a case of cutaneous bronchogenic cyst that occurred over the sternum in a 13-month-old boy.

CASE REPORT

A 13-month-old boy presented with a growing mass over the sternum that had been detected 2 weeks prior to admission. On physical examination, a non-movable, soft mass with inflammatory signs was detected on the overlying skin of the sternum. There was no sinus tract or discharge from the observed lesion. On ultrasonography, a well-circumscribed non-echogenic cystic mass, measuring $1.5 \times 1.3$ cm, was recognized in the subcutaneous fat layer over the sternum (Fig. 1). The excised cyst was unilocular and contained a yellowish seromucinous material. There was no connection between the cyst and the surrounding tissue. Histologically, the cyst was lined by ciliated, pseudostratified, columnar epithelium and goblet cells scattered among the ciliated epithelial cells (Fig. 2). There were several bundles of smooth muscle fibers (Fig. 3A) and mucous glands (Fig. 3B) in the cystic wall. There was histologic transition from columnar epithelium to stratified squamous epithelium in the lining epithelium (Fig. 4A). Numerous inflammatory cells, especially lymphocytes and plasma cells, infiltrating into the cystic wall to form lymphoid aggregates were present (Fig. 4B), and these cells were also infiltrated into the stratified squamous epithelium. Cartilage was not seen. Near the main cystic lesion, there was an incidental epidermal inclusion cyst having acute suppurative inflammation with a foreign body reaction. It had no connection with bronchogenic cyst.

DISCUSSION

Bronchogenic cysts are abnormalities of pulmonary differentiation that are usually detected in the pediatric patients. Patients with these lesions present with symptoms of infection or com-
pression on the vital structures. Most lesions are found in the mediastinum, along the tracheobronchial tree, or peripherally in the lung parenchyma. To date, 60 cases of cutaneous bronchogenic cyst have been reported in the English literature. Five cases of cutaneous bronchogenic cyst have been reported in Korean literature, and all of them have occurred in males.

Clinically, cutaneous bronchogenic cysts are cystic masses that are found shortly after birth or in early childhood. They are asymptomatic, and some of them have a fistulous opening that drains mucoid material. Males are predominant over females by almost four times. The most common location of cutaneous bronchogenic cyst is the suprasternal notch, followed by the presternal area,
The origin of bronchogenic cysts in the extrathoracic subcutaneous tissues can be readily explained by their embryologic development. The laryngotracheal groove separates the primitive foregut into dorsal and ventral structures beginning in the fifth week of gestation. In the seventh gestational week, this separation will be completed with a dorsal component forming the lung buds and a ventral component forming the foregut. Most bronchogenic cysts arise from the developing lung bud structures are formed during this period. Bronchogenic cysts in the subcutaneous tissue of the anterior chest wall occur either by anterior migration of an intrathoracic bronchogenic cyst or by a pinching off of the fusing sternal bars on the developing lung parenchyma. Extrathoracic cysts located in unusual sites (neck, shoulder and chin) can be explained by the migration of these sequestered structures in the developing embryo.

Cutaneous bronchogenic cysts are characterized by ciliated pseudostratified columnar epithelial cells interspersed with goblet cells, and these cysts also show other components including smooth muscle fibers, mucous glands and cartilages that are present in 80%, 53% and 7% of the cysts, respectively.

The following lesions should be considered in the differential diagnosis: cutaneous ciliated cyst, branchial cyst, dermoid cyst, and trichilemmal cyst. The definitive diagnosis of the excised lesion rests on the histologic evaluation. Bronchogenic cysts are typically lined by respiratory epithelium, and they often contain smooth muscle fibers, cartilages or mucous glands. Cysts of a branchial or thryglossal origin may contain respiratory epithelium, but smooth muscle fibers, cartilages or mucous glands should not be present. Cutaneous ciliated cysts occur in the lower extremities of female patients. The lining is ciliated columnar epithelium with papillary projections that resemble fallopian tubes. Thyroglossal duct cysts always appear as midline cervical masses. Their lining is ciliated columnar epithelium and they often contain smooth muscle fibers, cartilages or mucous glands should not be present. The lining of trichilemmal cyst is stratified squamous epithelium. Branchial cysts are lined by stratified squamous epithelium and surrounded by lymphoid tissue. Dermoid cysts are lined by an epidermis that possesses various epidermal appendages. The lining of trichilemmal cyst is squamous epithelium without an intervening granular cell layer.

In our case, the diagnosis may have been thrown into confusion because some of the histologic findings such as stratified squamous epithelium, and the lymphoid aggregates may suggest branchial cysts rather than cutaneous bronchogenic cysts. However, we considered the squamous epithelium as a metaplastic change due to the chronic inflammation. Lymphoid aggregates may be found in cutaneous bronchogenic cysts as was described by Beyer et al, who reported a case of a presternal bronchogenic anomaly with lymphoid aggregates. Unlike the cutaneous bronchogenic cysts, branchial cysts do not show smooth muscle fibers or secretory cells. Because of these reasons, we diagnosed this case as a cutaneous bronchogenic cyst.

Total excision is advised for the treatment of cutaneous bronchogenic cyst as mucocystic carcinoma can arise from a bronchogenic cyst, as was reported by Tanaka et al.

REFERENCES