

Case Report

Cyst in the hyoid bone: a case report and review of the literature

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Abstract: The most common congenital neck mass is the thyroglossal duct cyst. This cyst rarely occurs in the intrahyoid region. A review of the literature to date showed only five other reported cases of a cystic mass within the hyoid bone. We herein describe a 65-year-old woman who presented with a 4-year history of progressive swelling over the midline of the anterior neck. A computed tomography scan of the neck showed a benign-appearing cystic mass within the anterior portion of the mid-hyoid bone with well-defined margins. Under general anesthesia, the cystic lesion and central part of the hyoid bone were excised from the hyoid bone. The body of the hyoid bone (between the two lesser horns) and the remnant cyst were dissected away. Histological examination of the cyst showed cyst wall-like tissue. Given the radiographic, operative, and pathologic findings, the diagnosis of a thyroglossal duct cyst was confirmed. This intrahyoid localization once again supports a surgical approach to systematic resection of the body of the hyoid bone in patients with thyroglossal duct cysts.

Keywords: Hyoid bone, thyroglossal duct cyst, aneurysmal bone cyst, case report

Introduction

The most common congenital neck mass is the thyroglossal duct cyst (TDC), which accounts for 70% of all congenital neck anomalies [1]. However, localization of a TDC in the intrahyoid region is rare [2]. A review of the literature to date showed only five other reported cases of a cystic mass within the hyoid bone [2-6]. We herein describe a case of a TDC within the hyoid bone of a 65-year-old woman.

Case report

A 65-year-old woman presented with a 4-year history of progressive swelling over the midline of the anterior neck. The swelling had increased in size during the past month and had begun to cause pain and dysphagia. The patient had no significant medical or surgical history and denied any family history of thyroid disease or history of head and neck irradiation. On examination, a firm and non-tender swelling measuring 4 × 3 cm was found in the anterior neck at the level of the hyoid bone (**Figure 1A**). This mass was mobile with swallowing motions (pro-

trusion of the tongue) and did not infiltrate the skin. No associated cervical lymphadenopathy was present, and the rest of the physical examination was unremarkable. Laboratory test results were normal, and neck ultrasonography confirmed that the thyroid gland was in the normal position. A computed tomography scan of the neck showed a benign-appearing cystic mass within the anterior portion of the mid-hyoid bone with well-defined margins (**Figure 1B-D**). Chest radiography showed no abnormal findings.

Under general anesthesia, the cystic lesion and central part of the hyoid bone were excised from the hyoid bone (**Figure 2A, 2B**). A 5 cm transverse incision was made at the level of the hyoid. The muscles attached to the hyoid were elevated, and the hyoid bone was exposed at the midline, revealing an enlarged, thin-walled cyst. The anterior wall of the cyst was removed, and clear brown serous fluid was suctioned from the cavity. The cystic cavity was carefully examined, revealing no evidence of a tumor or mass inside. The body of the hyoid bone (between the two lesser horns) and the rem-

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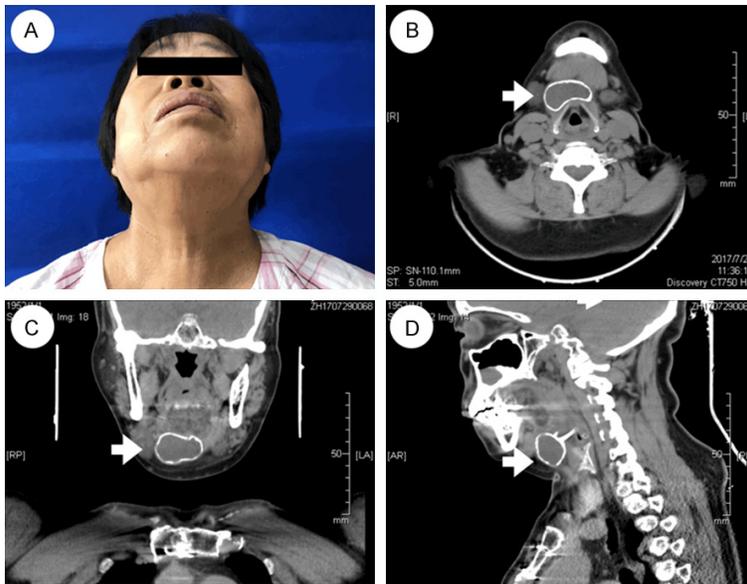


Figure 1. Physical and imaging examinations. A: Preoperative clinical view of lesion. B: Computed tomography, transverse view. C: Computed tomography, coronal view. D: Computed tomography, sagittal view.

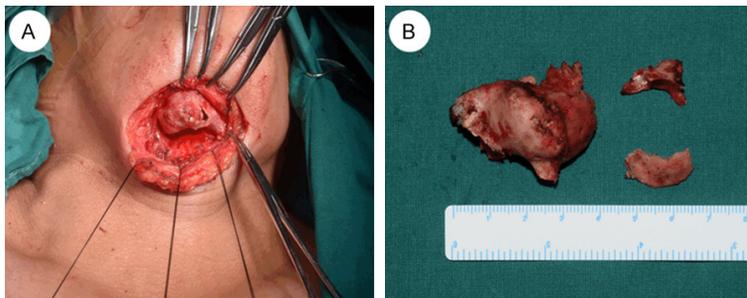


Figure 2. Gross anatomy of the lesion. A: Intraoperative view of hyoid bone cyst. B: Specimen in its entirety.

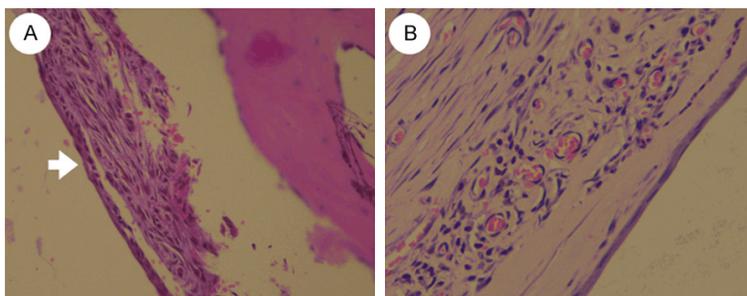


Figure 3. Histopathology of the resected lesion. A: The tissue comprised three layers: an inner cyst wall, osseous plate in the middle, and fibromuscular elements on the outside. The epithelial lining of the cyst wall was predominantly stratified squamous epithelium (arrowhead). B: Proliferation of fibrous connective tissue accompanied by chronic inflammatory cells was seen under the epithelial lining of the cyst wall (hematoxylin-eosin stain, original magnification $\times 200$).

nant cyst were dissected away. Postoperative recovery was uneventful. The patient had no complications during the postoperative period, and the drainage tube was removed on the second day after surgery. No disease recurrence was seen for 1 year postoperatively.

Histological examination of the cyst showed cyst wall-like tissue. The tissue was comprised of three layers: an inner cyst wall, osseous plate in the middle, and fibromuscular elements on the outside. The epithelial lining of the cyst wall was predominantly stratified squamous epithelium (**Figure 3A**). Proliferation of fibrous connective tissue was accompanied by chronic inflammatory cells, seen under the epithelial lining of the cyst wall (**Figure 3B**). No thyroid tissue or cholesterol granules were present in the cyst wall. Given the radiographic, operative, and pathologic findings, the diagnosis of a TDC was confirmed.

Discussion

We identified a few other reported cases of a cystic mass within the hyoid bone in the literature (**Table 1**) [2-6]. These midline cystic neck masses included TDCs, aneurysmal bone cysts, and unicameral or simple bone cysts.

The TDC is the most common congenital neck mass. It is derived from the persistence and dilatation of remnants of an epithelial tract that forms during migration of the thyroid during embryogenesis [7]. According to Shah *et al.* [8] TDCs are classified into four subdivi-

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Table 1. Summary of cases of cystic masses within the hyoid bone

References	Age/ Sex	Location	Presentation	Diagnosis	Gross Pathology	Treatment	Follow-up
Tas A <i>et al.</i> [3]	69/F	within the hyoid bone	painless and asymptomatic swelling	thyroglossal duct cyst of the hyoid bone	a mobile, palpable, asymptomatic and defined 3 × 5 cm mass	the cystic lesion and the thyroglossal duct were excised	uneventful; no recurrence of the disease
Bourjat P <i>et al.</i> [4]	67/F	in the body of the hyoid bone	reappeared, painless and asymptomatic swelling	thyroglossal duct cyst of the hyoid bone	a palpable, bilobular and well defined 55 × 25 × 35 mm mass	surgery (concrete method not mentioned)	not mentioned
Podoshin L <i>et al.</i> [2]	63/F	within the hyoid bone	painless and asymptomatic swelling	thyroglossal duct cyst of the hyoid bone	a palpable and well defined 4.5 × 2 × 2 cm mass	the mass and the hyoid bone were excised	uneventful
Shadaba A and Zaidi S [5]	18/M	through the body of the hyoid bone	progressive swelling associated with odynia and dysphagia	aneurysmal bone cyst of the hyoid bone	a palpable and defined 4 × 6 cm mass	total <i>en bloc</i> excision of the cyst	uneventful
Wang AS <i>et al.</i> [6]	60/M	involving the body of the hyoid bone	painless and asymptomatic swelling	unicameral bone cyst of the hyoid bone	a palpable and clearly defined 2.2 × 1.4 cm mass	transcervical marsupialization of the cyst	uneventful

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sions based on their location: (1) intralingual, (2) suprahyoid and/or submental, (3) thyrohyoid, and (4) suprasternal. Retrohyoid and especially intrahyoid localization is rare, and such TDCs are usually considered simple cysts attached to the periosteum. In our literature review, we found three cases of TDCs within the hyoid bone. Interestingly, these TDCs occurred in 63-, 67-, and 69-year-old women, similar to our patient. Tas *et al.* [3] and Podoshin *et al.* [2] reported cases in which the TDCs were completely within the hyoid bone, while Horisawa *et al.* [9] reported a case involving a 59-year-old man in which the TDC penetrated into the hyoid bone. A TDC within the hyoid is usually characterized by a lining of stratified squamous or respiratory pseudostratified ciliated columnar epithelium with mucus gland and thyroid follicles, meeting the criteria for the pathology of TDCs [10, 11]. However, Wei *et al.* [12] reported that 68 of 217 (31.3%) TDCs had ectopic thyroid tissue in the cystic wall. In another study, Chandra *et al.* [13] reported that thyroid follicles were identifiable in 34 (32.7%) of 104 TDCs. In the present case, no thyroid tissue was observed within the hyoid bone.

An aneurysmal bone cyst is a rare, non-neoplastic lesion that mostly involves the long bones and spine in young patients, and it is characterized by expansive, vascular, and multi-cystic features. Proposed etiologies include a benign neoplasm or a reactive lesion from trauma. Aneurysmal bone cysts of the head and neck are relatively rare, and when they occur, they are usually located in the mandible [14]. Shadaba and Zaidi [5] described a very rare case of an aneurysmal bone cyst within the hyoid bone. Computed tomography can define the lesion by its characteristic “soap bubble” appearance. Treatment options include surgical excision, curettage, intralesional injection of sclerosing agents, or arterial embolization.

A simple bone cyst is a benign fluid-filled cystic lytic lesion, which may be a unicameral bone cyst (UBC) or a partially separated bone cyst. UBCs can involve any bones, but they usually affect the metaphysis of long bones or the proximal humerus and proximal femur [15]. Other locations are rare or exceptional. Wang *et al.* [6] reported a case of a UBC within the hyoid. Histologically, UBCs are lined by fibrous connective tissue rather than epithelium. In the present case, the epithelial lining of the cyst wall was predominantly stratified squamous

epithelium; thus, the possibility of a UBC was ruled out. No consensus has been reached regarding the best treatment of UBCs, but treatment is generally unnecessary in asymptomatic individuals. UBCs can be treated with direct injection of a corticosteroid (methylprednisolone) into the cyst itself, curettage, or bone grafting [16]. Wang *et al.* [6] performed transcervical marsupialization of a UBC, achieving a therapeutic effect without recurrence.

The treatment of choice for TDCs is the Sistrunk procedure. Based on an anatomic and embryologic study, Dr. Sistrunk recommended removal of not only the cyst and central portion of the hyoid bone, but also a central core of deep tongue musculature [17]. In our case, considering that most of the central part of the hyoid bone was occupied by the cystic lesion and the patient developed pain and dysphagia, we resected the body of the hyoid bone. The cyst and tract remnant were not found in the muscles surrounding the hyoid bone during the operation. This intrahyoid localization once again supports the surgical approach to systematic resection of the body of the hyoid bone in patients with TDCs.

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Disclosure of conflict of interest

None.

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