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Case Report

Intrathyroidal branchial cleft cyst: a case report

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ABSTRACT

Intrathyroidal branchial cleft cyst is a rare entity in otolaryngology practice with few cases reported in the literature. Clinically, it mimics a thyroid swelling and the diagnosis is confirmed only by histopathology. The cyst walls are lined by predominantly non-keratinizing stratified squamous epithelium with characteristic subepithelial lymphoid tissue. This is a case report of right intrathyroidal branchial cleft cyst in a 50-year-old and a left intrathyroidal branchial cleft cyst in a twelve- year-old boy. Both patients underwent imaging followed by surgical management. A fourth branchial cleft cyst is very rare and may have varied presentation. Both patients with intrathyroidal branchial cleft cysts had complete surgical excision and are normal during regular follow ups. The presentation of a fourth branchial cleft cyst may occur as an intrathyroidal swelling. The recommended treatment of an intrathyroidal branchial cleft cyst is complete surgical excision of the branchial cyst with thyroidectomy. Misdiagnosis of a fourth branchial cyst can result in incomplete removal leading to recurrence. Awareness about the clinical manifestations of a fourth branchial anomaly is crucial for appropriate management.

Keywords: Intrathyroidal branchial cleft cyst, Thyroid swelling, Surgical excision

INTRODUCTION

Branchial anomalies are congenital anomalies that are derived from the branchial cleft apparatus that persists after fetal development.[1] A branchial cleft cyst (BCC) results from the failure of obliteration of the cervical sinus.[2] A BCC is an epithelial lined structure without an external opening and it is commonly located in the lateral aspect of the neck.[3] Cysts with similar histological features of branchial cleft cysts have been seen in unusual sites, such as oral cavity, parotid, pancreas, and thyroid.[4]

An intrathyroidal branchial cleft cyst is a rare entity which should be considered in the differential diagnosis of a thyroid lesion. Branchial cleft remnants may exist de novo within the normal thyroid gland which suggests that the branchial clefts may contribute towards normal thyroid gland elements as well as to parafollicular C cells.

During fine needle aspiration of a thyroid cyst, if pus-like material is aspirated, the possibility of intrathyroidal branchial cleft cyst should be considered.[1] The treatment is complete surgical excision and the diagnosis is clinched by histopathology.

Cysts of branchial cleft origin have been reported to have a squamous epithelial lining and abundant lymphoid tissue with reactive germinal centers on histopathology.[5]

CASE REPORTS

This is a case report of two patients with intrathyroidal branchial cleft cyst which is a rare occurrence.

Case 1

A 50-year-old lady presented with a neck swelling of two-years duration. Examination revealed a smooth mass measuring about 45×25×15 mm in the right thyroid lobe which was moving up with deglutition. There was no mediastinal extension. Sonography and computed tomography showed a cystic lesion with solid component in the right thyroid lobe (Figure 1). Thyroid function evaluation and other laboratory tests were within normal limits. To rule out the possibility of thyroid malignancy, the patient underwent fine needle aspiration of the lesion. The cystic component of the lesion in the right half of the thyroid gland was also aspirated. The aspirated fluid measured about 1 ml and was thick yellowish pus-like fluid showing features suggestive of thyroid tissue. Complete excision of the cyst with right hemithyroidectomy was performed. Histopathological examination showed thyroid tissue with a cyst lined predominantly by ciliated columnar epithelium and focally by squamous epithelium with lymphoid aggregates with no evidence of malignancy. The features were consistent with a branchial cleft cyst (Figure 2). The patient healed well without any recurrence and is on regular follow up.

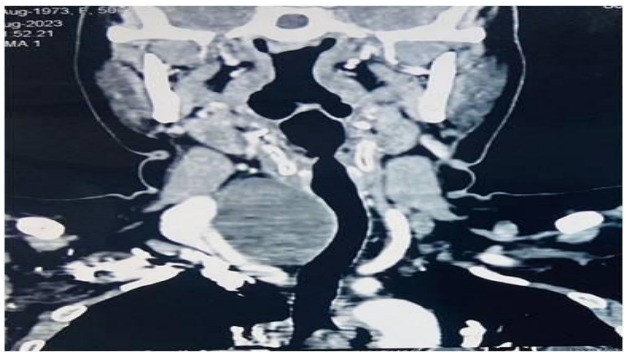

Figure 1: Large right thyroid nodule seen on coronal CT scan.

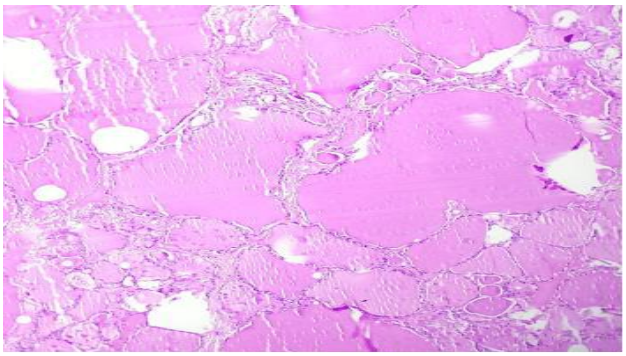

Figure 2: Histopathologic features consistent with an intrathyroidal branchial cyst.

Case 2

A 12-year-old boy presented with complaints of swelling in the lower left side of the neck with discharge for several years. There was a history of incision and drainage done elsewhere in the past. Examination revealed a left thyroid swelling with discharging sinus and scar tissue. Ultrasound neck showed a sinus tract with skin opening and mobile internal echoes visualized in the left side of the neck with external opening. The sinus tract was seen passing along the posterior aspect of the left lobe of thyroid gland up to the lateral wall of trachea. An enlarged level two lymph node was seen. Computed tomography (CT) neck plain and contrast as well as a sinogram showed a sinus tract with skin opening at the left side of the lower neck medial to the sternocleidomastoid muscle at the level of the thoracic inlet with subcutaneous cystic area and surrounding edema coursing posteriorly into the defect in the left lobe of thyroid and posterosuperiorly along the posterior aspect of the left lobe of thyroid possibly up to the left pyriform sinus (Figures 3-5). Sinogram showed opacification of the sinus tract in the subcutaneous soft tissue up to the lateral margin of the left lobe of thyroid gland measuring 1.8×0.9 cm. The length of the sinus tract up to the upper pole of left lobe of thyroid measured 4.5 cm. Excision of the cyst along with partial hemithyroidectomy was done (Figures 6 and 7). The thyroid cyst was dissected including the tract which was traced up to the pyriform sinus and completely removed. Histopathology was reported as colloid nodule with adjacent cystic tract consistent with branchial cyst (Figure 8).

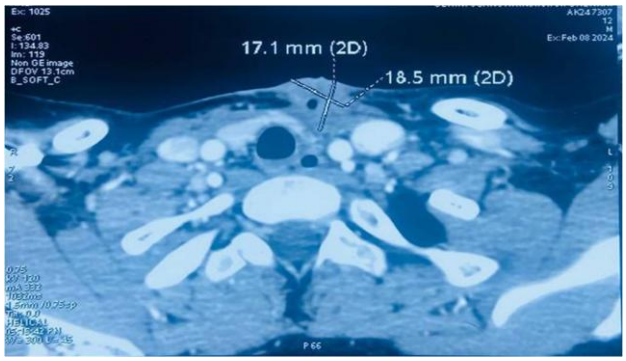

Figure 3: CT scans axial view shows a cystic thyroid swelling on the left side.

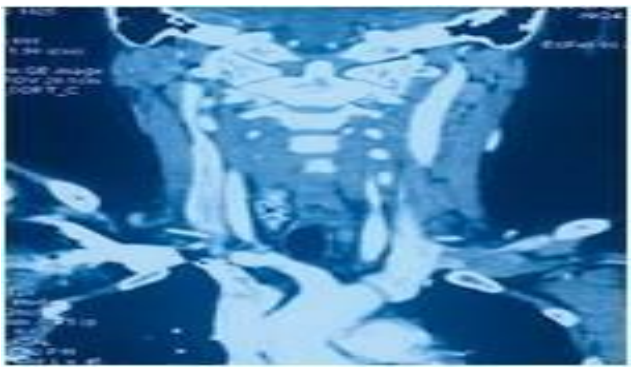

Figure 4: CT scans sagittal view shows the tract of the branchial cyst coursing posterosuperiorly.

The 50-year-old lady who presented with a thyroid swelling underwent pre-operative imaging and fine needle aspiration followed by complete excision of the cyst with right hemithyroidectomy. The patient is on regular follow up for 18 months and has no recurrence. The 12-year-old boy with a left thyroid swelling with a discharging sinus underwent ultrasound neck, CT neck plain and contrast as well as a sinogram pre-operatively. He underwent complete excision of the cyst along with left hemithyroidectomy. He is doing well with no evidence of disease and is on follow up for 13 months.

Figure 5: CT scans sagittal view shows the tract of the branchial cyst coursing posterosuperiorly possibly up to the left pyriform sinus.

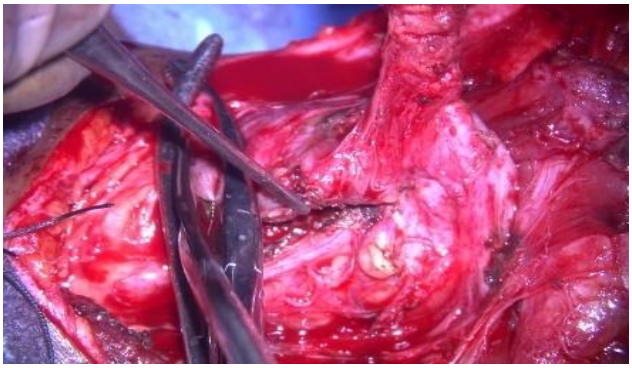

Figure 6: Branchial cyst with tract dissected along with surrounding thyroid tissue from left lobe of thyroid.

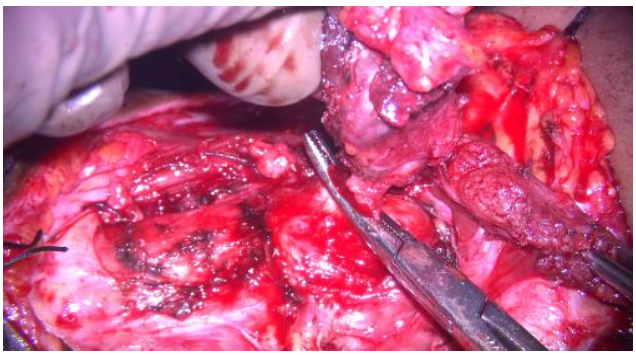

Figure 7: Branchial cyst with track traced upto the left pyriform sinus and completely excised.

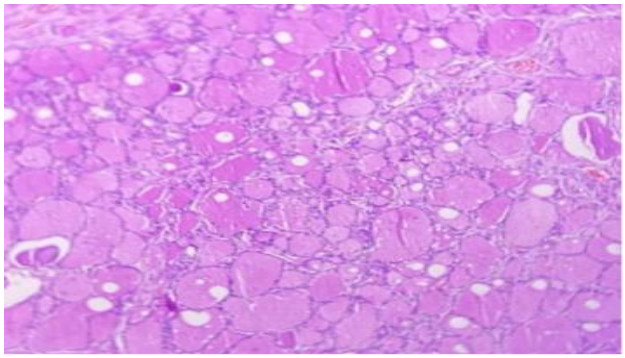

Figure 8: Histopathology was reported as colloid nodule with adjacent cystic tract consistent with branchial cyst.

DISCUSSION

Branchial cleft anomalies are uncommon congenital anomalies encountered by the otolaryngologist. The branchial apparatus develops during the fourth week of gestation and consists of a series of arches, pouches, and grooves which extend from the developing oral cavity to the respiratory diverticulum. The term 'branchia' is greek for gill, and reflects the resemblance of this human embryological structure to the gills seen in fish. Abnormal persistence of branchial apparatus remnants results in branchial anomalies.[3] Branchial anomalies have traditionally been classified as cysts, sinuses, and fistulas.[1] von Baer in 1827 gave the earliest description of the branchial apparatus and von Ascherson in 1832 is credited with the earliest recognition of branchial cleft anomalies.[3] Branchial cleft cysts attributed to the persistence of branchial remnants have been recognized since the 18th century.[4] Branchial cleft cysts are noted in the lateral aspect of the neck, predominantly occurring in the fourth decade of life and without sexual predominance.[6] Although branchial cleft cysts are common in the lateral aspect of the neck, cysts with similar histologic features of branchial cysts have been reported in unusual locations e.g. thymus, oral cavity, parotid gland, pancreas and thyroid.[7] Branchial cysts originate from entrapped remnants of branchial clefts or pouches or of persistent ectoderm of the cervical sinus.[3]

Fourth branchial arch anomalies represent 1% to 2% of all branchial anomalies.[3] The presentation is most common in the pediatric age group although adults may also present with this entity. Fourth branchial cleft cysts can be seen anywhere in the neck down to the mediastinum, although they are mostly seen adjacent to the thyroid gland. Louis et al. were the first to report on the occurrence of branchial cleft cysts within the thyroid gland in 1987.[8] Branchial cleft remnants exist de novo within the normal thyroid gland and this suggests that the branchial clefts may contribute towards normal thyroid gland follicular cells as well as to parafollicular C cells.[9] The exact histogenesis of an intrathyroidal branchial cleft cyst is not known. The failure of the third or fourth branchial pouches to obliterate

in utero results in the formation of cysts or sinus tracts that lie close to, or inside the thyroid gland.[1]

BCC may present as thyroid masses and may be partly or completely intrathyroid. A history of recurrent upper respiratory tract infection, neck or thyroid pain/tenderness, as well as neck mass is common. The external sinus tract opens at the lower lateral neck below the thyroid and cricoid cartilage anterior to the sternocleidomastoid muscle. The fistula starts from the apex of the pyriform sinus and emerges between the thyroid and cricoid cartilages cranial to the recurrent laryngeal nerve and caudal to the superior laryngeal nerve.[10] At times, a sinus tract may not be present, hence there is no external opening. Other presentations of BCC include cellulitis, hoarseness, odynophagia, thyroiditis, abscess, and rarely stridor. The link between acute suppurative thyroiditis and pharyngeal pouch remnants was initially reported in 1979. Thyroid abscesses in children often indicate an underlying branchial remnant, especially when cultures reveal a mixed flora.[11] Chronic thyroiditis may act as a trigger for squamous metaplasia, and enlargement of these metaplastic foci can result in the formation of squamous epithelial lined cyst.[4] The presence of squamous epithelium is not usual within the thyroid gland.[7] Malignant transformation of BCC is extremely rare.[12]

It is important to distinguish between intrathyroidal branchial cleft cyst and other thyroid lesions with squamous epithelium, such as congenital remnants, benign metaplasia, and malignancy.[8] Squamous epithelium in a thyroid cyst may be derived from thymic remnants, thyroglossal duct remnants, metaplastic follicular cells, ultimobranchial remnants, and tumors containing squamous cells.[7] Thyroglossal duct cysts, cystic thymic remnants, and epidermal inclusion cysts are examples of congenital remnants.[8] Thyroglossal duct cyst has walls composed of alternating non-keratinizing stratified squamous epithelium and respiratory epithelium, while although respiratory epithelium may be seen in the wall of a branchial cleft cyst, its presence is minimal. Normal thyroid tissue is typically found with thyroglossal duct cyst.[2] Squamous cells may also be found in thyroid glands involved by inflammation. The formation of branchial-cleft-like cysts in the thyroid is reported to be associated with Hashimoto's thyroiditis.[7] Delabie et al. reported that BCC probably originate from remnants of the ultimobranchial body (also known as solid cell nests). Papillary carcinoma, mucoepidermoid carcinoma, squamous carcinoma, adeno-squamous carcinoma, and teratoma are thyroid tumors that can display squamous metaplasia.[8]

On ultrasound, the branchial cleft cyst is reported to appear as an anechoic mass or a predominantly hypoechoic, cystic mass with faint internal debris and posterior enhancement. Occasionally, the BCC may be hyperechoic with a pseudosolid appearance. Fine-needle aspiration of yellowish, green viscous fluid or the ultrasonogram appearance of a pseudosolid, cystic nodule in the thyroid gland suggests a congenital developmental cyst.[1] Low-density, well-defined unilocular mass with thin uniform enhancing rim is typically seen on CT scans.[6]

Histologically, most branchial cysts are reported to be lined by squamous epithelium, although some are lined by respiratory (pseudostratified ciliated columnar) epithelium, and others are lined by both squamous and respiratory epithelium. The cysts usually possess a considerable amount of subepithelial lymphoid tissue, often in a follicular pattern with germinal centers. Occasionally, sebaceous glands and salivary tissue are known to be present.[3]

The cystic contents have been reported to be clear, watery to mucinous fluid, the cyst may contain desquamated, granular cellular debris; when infected, it may be yellowish pus-like fluid.[1] In the epithelial lining of a branchial cleft cyst, positive immunohistochemistry (IHC) staining with galectin 3, high molecular weight cytokeratin (HMWCK), and localized staining with carcinoembryonic antigen (CEA) has been reported.[8] Even if a branchial cleft cyst has been diagnosed by histopathology, there may be concurrent thyroid papillary carcinoma.[6] A rare case of bilateral intrathyroidal branchial cleft cysts in a 2-day-old neonate who presented with intermittent tachypnea, dyspnea, and a palpable neck mass has been reported.[13]

The possibility of a branchial origin for a thyroid swelling must be kept in mind as complete excision including the tract will prevent recurrence. In our experience, one adult and one pediatric patient were found to have intrathyroidal branchial cyst. Complete excision of the cyst and the tract with partial thyroidectomy helped ensure a successful outcome. Both patients are under periodic review and have had no recurrence over more than a one-year follow-up. In both our patients, the suspicion of a branchial origin was thought about pre-operatively and hence the cyst with the tract was completely removed.

The limitation of this case report is the experience with a limited number of patients (two patients) and lack of a long term follow up.

CONCLUSION

Intrathyroidal branchial cleft cyst is an uncommon lesion in otolaryngology practice. Embryonic remnants of the fourth branchial pouch give rise to fourth branchial cleft cysts. On histopathology, the cyst is usually lined by squamous epithelium and mucous cells, containing abundant lymphoid tissue with germinal centers. Awareness of the possibility of a branchial origin for a thyroid cyst is important for complete excision and avoidance of recurrence.

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