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Papillary Thyroid Carcinoma Arising in Thyroglossal Duct Cyst: A Case Report

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ABSTRACT

Thyroglossal duct cysts (TGDC) are usually located in the midline of the neck. A remnant of the thyroglossal duct, usually a cyst, is the most common congenital abnormality of thyroid gland development. This study aimed to describe a patient with TGDC and the microscopic appearance of the supporting examination. A woman, 22 years old, was admitted to the hospital with complaints of a lump in the submandibular area. Excision surgery was performed, with the result microscopic showing the cyst was composed of cuboidal epithelial cells and thyroid follicles present in the cyst wall, and some histological findings, such as; formation of papillary structures, nuclear morphologic variations such as ground glass nuclei, pseudoinclusions, intranuclear grooves. Based on the clinical symptoms and histopathology, the mass in the left submandibular is consistent with papillary carcinoma arising in thyroglossal duct cysts.

1. Introduction

The thyroglossal tract is a tract epithelium formed from the thyroid gland that descends from the foramen caecum, normally located in the thyroid cartilage, and will disappear at 5 to 10 weeks of gestation. Thyroglossal duct cysts (TGDC) is formed from incomplete atrophy of the thyroglossal tract or persistent epithelial cysts. Remnants of the thyroglossal tract may be cysts, tracts, ducts, or ectopic thyroid cysts or ducts.[1,2] Primary malignancy in TGDC is a very rare case, and 0.7-1% of cases have been reported. Papillary carcinoma was found in 80-85% of cases and is the most common carcinoma in TGDC, followed by squamous cell carcinoma.[1,2] This study aimed to describe a patient with TGDC and the microscopic appearance of the supporting examination.

2. Case Presentation

A woman, 22 years old, was treated at the hospital with the complaint of a lump on the left side of the neck, which has been getting bigger with intermittent pain for 1 year. There was no history of hypertension nor diabetes mellitus, or asthma/allergy. Physical examination showed vital signs within normal limits. In the neck area, there is a lump in the left submandibular region, size 10x 8x 6 cm. The boundary with the surrounding area is not clear. On

palpation, the consistency is firm and fixated. No bruit on auscultation. On supporting examinations with x-rays and ultrasonography, the impression of a benign cyst was found.

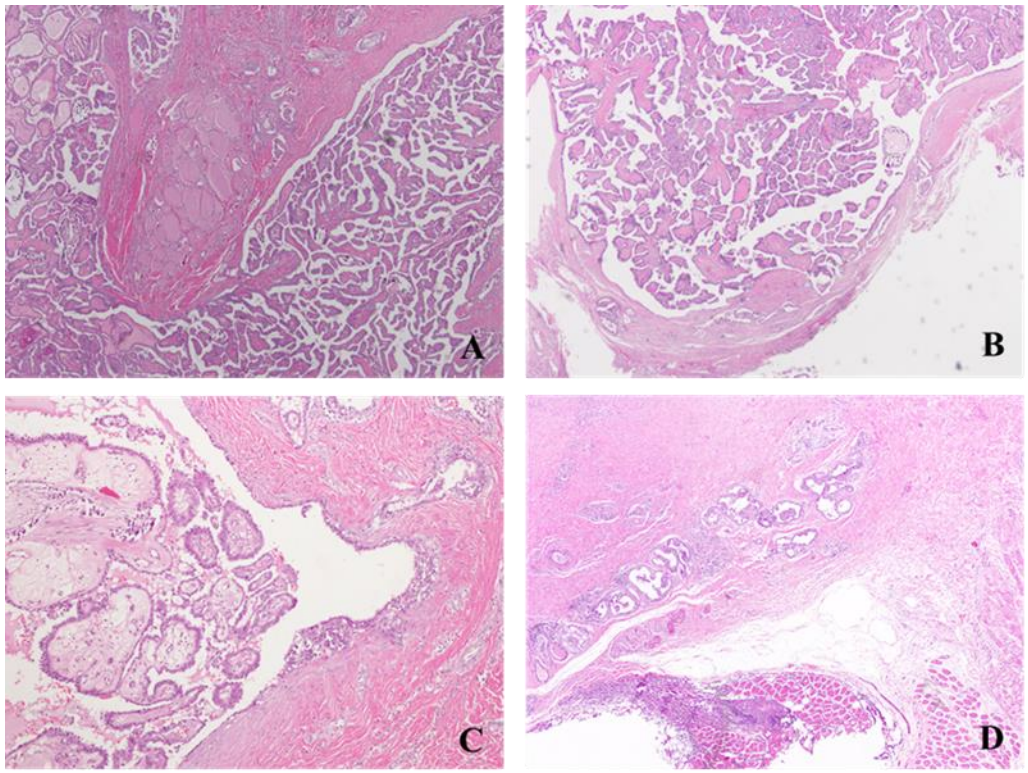

Figure 1. Morphology of papillary structures in the lumen of cyst. (A) The cyst is lined by a single layer of cuboidal epithelium, the cyst wall is made of fibro collagenous connective tissue, and benign thyroid tissue (colloid-filled follicles) within the cyst were still noticed. (B) The growth of the tumor with papillary fronds with fibrovascular stalk. (C) The growth of the tumor with a pseudo follicular pattern. (D) This figure showed invasive papillary thyroid carcinoma within a thyroglossal duct cyst with the extension of the tumor into the strap muscles.

Excision was conducted, and a microscopic examination exhibited a cyst structure lined by cuboidal epithelium. On one region close to the cystic wall, thyroid follicles of varying shape and size are found, lined with a simple cuboidal epithelium with colloid in the lumen (Figure 1A). Inside the cyst was filled with full masses with papillary and micropapillary structures covered with neoplastic cells with single or pseudostratified nuclei, the majority of which were cuboidal in shape, round, oval nucleus, vesicular, some with longitudinal groove, nuclear pseudo inclusion (Figure 1 A-D). Ground glass appearance, eosinophilic cytoplasm. Psamomma bodies were found between the tumor group and the stroma of fibrous connective tissue (Figure 2). The stroma is also filled with inflammatory cells of lymphocytes, plasma, hemosiderophages, and the proliferation of blood vessels.

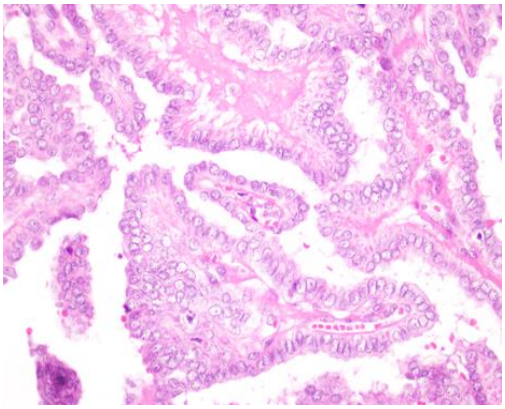

Figure 2. Characteristics papillary thyroid carcinoma. At higher magnification, nuclear features diagnostic papillary thyroid carcinoma were present, showing overcrowding of nuclei with nuclei grooving and intranuclear pseudoinclusion.

3. Discussion

Thyroglossal duct cystic (TGDC) is a congenital mass that is often found on the neck. This cyst is a persistent cystic dilatation of the thyroglossal duct, which is located in the midline of the neck, above the thyroid isthmus, and below the hyoid bone. In the process of development, the thyroid gland descends to where it should be through a duct called the thyroglossal. Normally this duct will involute. The patency of this duct gives rise to a great potential for the formation of a thyroglossal duct cyst. The coexistence of carcinoma in thyroglossal duct cysts is extremely rare, with most being papillary carcinoma. In this case, there was no difference in age, and generally, they were found before the age of the 4th decade. It was more common in children, with a predilection for the age of 0-20 years (52%) and 38% for ages up to 5 years. Sistrunk reported 31 cases out of 86,000 pediatric patients. This case is very rarely found in the lateral part of the midline, for example, the lateral jugular vein.[3]

Clinically, most thyroglossal duct cysts are benign and present as slow-growing, asymptomatic neck masses. On physical examination, the mass is found along the midline, mobile, and located at the hyoid bone or below. The mass is very rarely found in suprahyoid, submental, intrathyroidal, or even intralingual. The cyst may communicate with the skin or the foramen caecum of the sinus tract. It also moves up when swallowing, and if there is an infection, the pain will accompany it. Most cases are asymptomatic, and in as many as 10-20% of cases, fistula, sinus draining, and dysphagia are found, while there is very rarely airway obstruction. Cysts can fluctuate in size, especially if infection occurs. In neonates, airway compression can occur with symptoms of apnoea, cyanosis, and respiratory problems.[3-6]

There are some useful methods of carcinoma in TGDC diagnosis, but none of them is applied in all cases. The importance of CT and TC scans in thyroid diagnosis is underlined. However, they are advisable only in patients with either abnormal thyroid function tests or the absence of the thyroid gland proper during ultrasonography. Some authors strongly recommend FNAB as a safe, well-tolerated, and cost-effective procedure in diagnosing thyroglossal duct lesions. Nevertheless, cytology smears are inconclusive in approximately 50% of the described cases. The major diagnostic criteria for papillary carcinoma are high cellularity, the presence of papillary formations, and enlarged nuclei with powdery chromatin and with definitive nucleoli.[3-5]

Macroscopic tumor mass is usually less than 2 cm, with a smooth cyst surface. Microscopically, the cyst is lined by columnar or squamous epithelium in

60% of cases. Thyroid tissue can be found to be hyperplastic, nodular, neoplastic, or normal. If inflammation is found, squamous metaplasia can be found.[3,4] For the development of a thyroglossal duct cyst to become a carcinoma is very rare, especially in the form of papillary thyroid carcinoma, which can be diagnosed during fine needle aspiration based on the presence of papillary structures with individual nuclear grooves or intranuclear inclusions. The presence of papillary thyroid carcinoma (PTC) in TGDC should exclude a primary thyroid tumor.[3,4] Another variant can be found, such as squamous (epidermoid) carcinoma. While medullary carcinoma is associated with C cells, therefore, it cannot be found in TGDC. Follicular cells are embryologically different. The C cells develop from the ultimobranchial apparatus through the neural crest and migrate to the lateral thyroid but not to the midline.[3]

The differential diagnosis for TGDC are branchial cleft cyst and cervical thymic cyst, where the location is generally lateral, no thyroid follicle is found, and it is not associated with the hyoid bone. Metastatic (cystic) PTC is distinguished by clinical features and radiological evaluation. It was found that there was a transition of TGDC cells to PTC.[4] Prognosis of TGDC papillary thyroid carcinoma seems to be similar to that of papillary carcinoma of the thyroid gland. Papillary thyroid carcinoma has a good prognosis. Metastases only occur in 2% of cases, while squamous cell carcinoma has a poor prognosis. The incidence of cervical metastases is very low, as reported in 8% and 1.3% of cases.[7-9] two theories explain the thermogenic origin of TGD adenocarcinoma. First, de novo theory in 62% of cases, ectopic thyroid tissue can be found microscopically, and no medullary carcinoma originating from parafollicular cells is found. The second metastatic theory states that thyroglossal cyst carcinoma originates from the primary thyroid gland, namely papillary carcinoma.[5,8]

Regarding the management, some authors believe that papillary carcinoma thyroid arising in TGDC, without cervical metastasis, and if a radiologically normal thyroid gland is found, it can be managed adequately by Sistrunk's operation, thyroid suppression, and strict long-term follow-up.[2] The Sistrunk procedure includes resection of the entire thyroglossal duct with a cyst or fistula and 1-2 cm from the center of the hyoid bone. If the hyoid bone segment is not removed, the recurrence rate is above 25%. The well-differentiated form of carcinoma that arises from TGDC has a good prognosis with the Sistrunk procedure. On the other hand, other authors have suggested a more aggressive approach characterized by the Sistrunk procedure, total thyroidectomy. Post-operative radioactive iodine therapy and thyroid hormone replacement, based upon the observation that papillary thyroid carcinoma may metastasize through the thyroglossal duct remnant without a lesion being clinically detected in the gland itself.[5-8,10]

4. Conclusion

There are many controversies about the origin of the tumor and the extension of surgery needed, making it difficult to define many aspects related to its management and follow-up.

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