Primary Papillary Thyroid Carcinoma in The Thyroglossal Cyst

Over Eighty Months Follow Up

Mehmet Akif Eryılmaz, Hatice Toy

Selçuk University, Medical Faculty, Departments of Otolaringiology and Pathology, Konya, Turkey

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***Correspondence**: Mehmet Akif Eryılmaz*
Selçuk Üniversitesi Meram Tıp Fakültesi
Göğüs Hastalıkları AD. Konya
E-mail: akiferyilmaz@hotmail.com

ABSTRACT

Papillary thyroid carcinoma within the thyroglossal duct cyst is a very rare finding and its presentation is like the benign cyst. A 22 years old female patient with a midline neck mass was initially diagnosed thyroglossal duct cyst and underwent Sistrunks procedure and upon histopathologic examination she was diagnosed papillary thyroid carcinoma within the thyroglossal cyst with regional lymph node metastasis. Than she sent to radioactive iodine ablation treatment after the thyroid gland evaluation and she was followed up over eighty months free of disease. Sistrunk procedure is usually treatment of choice for the thyroglossal duct cyst, but we could rarely find malignant thyroid carcinoma within the cyst. Than we should reevaluate the thyroid gland and if there is a positive finding we should add some additional treatment like radioactive iodine ablation, total thyroidectomy, and neck dissection.

Key words: *Papillary, thyroid, carcinoma, thyroglossal duct cyst*

Tiroglossal Ductus Kisti İçerisinde Primer Tiroid Papiller Karsinomu Ve Seksen Aydan Fazla Takibi

Tiroglossal duktus kisti içerisinde primer tiroid papiller karsinomu çok nadir bir durumdur ve benin bir kist gibi bulgular verir. Boyun orta hatta kitlesi olan 22 yaşında bayan hasta başlangıçta tiroglossal duktus kisti tanısıyla Sistrunk ameliyatı oldu, histopatolojik inceleme sonucunda tiroglossal duktus kisti içinde primer tiroid papiller karsinoma ve bölgesel lenf nodu metastazı belirlendi.Tiroid bezi incelemelerinde hastalık tespit edilmeyen ve sonrasında tamamlayıcı cerrahiyi kabul etmeyen hastaya radyoaktif iyot ile ablasyon yapıldı ve seksen aydan uzun bir süredir ilave hastalık belirtisi olmadan takip edildi. Tiroglossal duktus kistlerinde Sistrunk ameliyatı tedavi seçeneğidir ancak histopatolojik inceleme sonucunda tiroid papiller karsinomu tespit edilirse tiroid bezi ve çevre dokular tekrar incelenerek pozitif bulgu varsa total tiroidektomi ve boyun diseksiyonu, radyoaktif iyot ile ablasyon tedavisi gibi tedaviler eklenmelidir.

Anahtar kelimeler: *Papiller, tiroid, karsinom, tiroglossal duktus kisti*

INTRODUCTION

Thyroglossal ductus (TGD) remnants are the most common midline neck mass usually located at the level of the thyrohyoid membrane (1). The possibility of malignancy within the thyroglossal duct remnants are 1-2% (2). There are over 215 primary thyroid malignancies in the thyroglossal ductus remnants in the literature. Most of the malignancies are histologically papillary carsinomas (80%); there are also few cases of follicular, squamous cell and other types of carsinomas (3). In most of the cases the diagnosis of the thyroglossal ductus carcinoma are made after the histopathologic examination of the surgical specimen of a routine Sistrunk procedure (4). The typical patient is a woman who is 20 to 60 years of age with a midline servical swelling at the level of the thyrohyoid membrane without pain, tenderness (5). Hence the diagnosis of the carcinoma is usually made after the surgical procedure, there is a debate if the Sistrunk procedure is enough or should we add some other treatment modalities like thyroid hormone supplementation to reduce thyroid stimulating hormone (TSH), radioactive iodine ablation, total thyroidectomy for occult carcinoma in thyroid gland or neck dissection for the occult lymph node metastasis (4). In this study we report a case of primary papillary thyroid carcinoma within the thyroglossal ductus cyst with regional lymph node metastasis and over eighty months of follow up of the patient.

CASE

A 22 years old female patient admitted to the Konya Numune Hospital ENT outpatient clinic with the chief complaint of a cervical mass, otherwise healthy. She realized the mass six months ago. Physical examination revealed non tender 2.5x1.5 cm midline cervical mass at the thyrohyoid membrane level. The mass was smooth, mobile, elastic and painless by palpation. There were no other palpable lymph nodes and the thyroid gland was nonpalpable. Thyroid hormone levels were within normal limits. On ultrasonographic examination the thyroid glands dimensions were normal and paranchyma echogenicity was homogenius and there were no nodules. There were no cervical lymph nodes. There was a midline 2x1 cm diameter neck mass slightly hypoechogenic superior to thyroid isthmus with smooth shape. The patient underwent a Sistrunk procedure. During the procedure the dissection of the cystic mass up to the hyoid bone was carried out easily.

The histopathologic report was macroscopically 1.5x1.1 cm grayish white colored noduler soft tissue and 5x0.4 cm bony tissue. Microscopically thyroid papillary carcinoma within the thyroglossal ductus cyst and two micro metastatic lymph nodes adjacent to the specimen were reported. In Figure 1, histology section showing the cyst wall and the tumor (H&E x 10). In Figure 2, histology section showing the papillary thyroid carcinoma characteristic futures like ground glass appereance, papillary structure (H&E x 20). Then the patient was reevaluated for a possible thyroid disease and neck metastasis. Thyroid function tests, ultrasonography and scintigraphic scan revealed that the thyroid gland and the neck were free of the disease. The patient was sent to radioactive iodine ablation treatment because of the positive regional lymph nodes. She is also taking levothyroxin for suppression of the TSH. More than eighty months follow up revealed no sign of disease.

DISCUSSION

The treatment of the most common midline cervical mass is the Sistrunk procedure in which the mass and the canal is carefully dissected with the body of the hyoid bone up to the foramen cecum (6). During embriogenesis the thyroid gland first appears at the beginning of the aerodigestive tract and migrates to the adult location. The remnants of the canal could be any where between foramen cecum and thyroid gland. There could be thyroid follicles within the ductus and cyst.

Plaza CP et al. in their five cases series recommends Sistrunk procedure and total thyroidectomy in selected cases. Their patients tumor sizes were 15 to 40 mm. they also performed bilateral neck dissection in one patient (4). Weiss and Orlich in their study found 11.4% TGD cyst carcinoma and microscopic carcinoma foci in the thyroid gland. They recommended total thyroidectomy following the histopathological diagnosis of papillary carcinoma in the TGD cyst, despite the thyroid gland was normal in the initial assessment (7).

On the other hand, Kojima et al. did not recommended total thyroidectomy in cases of clinical and intraoperative diagnosis of a normal thyroid gland (8). Patel et al. did not recommended additional treatments in low-risk patients (5). Patients below 45 years old of age, with tumor diameter <4 cm, and no sign of extensive metastasis were low risk (9). Our patient was 22 and tumor diameter was 1.5 cm, thus categorized as low-risk. Patel

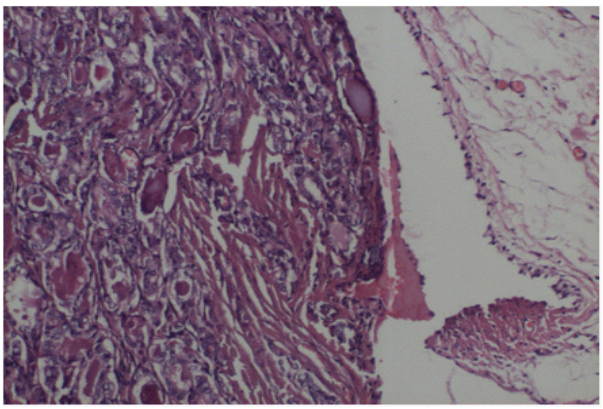

***Figure 1**. Histology section showing the cyst wall and the tumor (H&E x 10).*

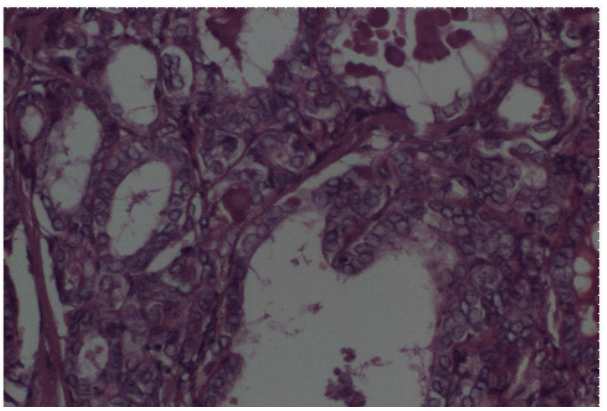

***Figure 2**. Histology section showing the papillary thyroid carcinoma characteristic futures like ground glass appereance, papillary structure (H&E x 20).*

et al and Chu et al. also rejected to made thyroidectomy when pathological findings were not present in the preoperative ultrasonographical and biochemical examinations and additional clinical and pathological findings were absent (5,10). Our patient was initially diagnosed as TGD cyst, she underwent a Sistrunk operation. In her pathologic report we realized that she had a malignant disease than we reavaluated her for a possible thyroid disease but there were no positive results. We sent the patient to radioiodine ablation because of the positive regional lymph nodes. After eighty months we reevaluated the patient for a possible thyroid disease but there was no sign.

In conclusion, there are very limited cases cases of the primary TGD cyst carcinomas and more limited follow up reports in the literature. We should consider a possible malignancy in the TGD cyst before a Sistrunk procedure although its very rare. When we diagnose a TGD remnant we should strongly recommend the patient a Sistrunk procedure because the possibility of the malignancy increases by age. We also should evaluate the thyroid gland for a possible malignancy.

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