Papillary carcinoma of thyroglossal cyst

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Abstract

Malignancy of thyroglossal cyst is a rare entity. Medical literature has reported only a very few cases till date. Histopathologically papillary carcinoma of thyroglossal cyst is the most common variety encountered in clinical practice. Surgical excision by Sistrunk operation is the standard treatment of choice followed by subtotal or total thyroidectomy whenever indicated. We report here two cases of thyroglossal cyst carcinomas and their respective management.

Keywords Thyroglossal cyst · Papillary carcinoma

Introduction

Thyroglossal cyst is a benign clinical condition second only to cervical lymph node as the most common cause of solitary midline neck swelling [1, 2]. Neoplastic change within the thyroglossal cyst is however rare and only a few cases have been reported in medical literature. Recently we have encountered two cases of thyroglossal cysts, which turned out to be cases of papillary carcinoma, one of which being unusually large. We discuss the two cases and the various therapeutic options herewith.

Case history

Case 1

A 26-year-old female patient presented to Otolaryngology clinic with history of a midline neck swelling for three years, which had been progressively increasing in size for the last six months.

On clinical examination the mass was 6 x 3 cm in dimension, situated strictly in midline, moved on deglutition as well as on protrusion of tongue (Fig. 1). The mass was tender on palpation, firm to hard in consistency, non pulsatile and not fixed to any underlying structures. There was no evidence of any sinus or fistula. Clinical examination of neck did not otherwise reveal any abnormality.

Routine hematological and biochemical investigations were found to be within normal limits. USG guided FNAC revealed it to be a case of thyroglossal cyst. USG of thyroid gland as well as pre operative thyroid scan were found to be within normal limits.

The mass was excised under general anaesthesia by Sistrunk operation. It was subhyoid in location and had extensive adhesions to the surrounding structures. The central part of body of hyoid bone along with the thyroglossal tract upto foramen caecum was excised. Per operative examination of the thyroid gland was within normal limits. As the

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Fig. 1 Photograph of the patient during clinical examination with the mass in midline.

Fig. 2 Photograph of the mass after removal.

Fig. 3 Photograph of the histopathological slide (10x 10 magnification) showing the cyst wall containing thyroid follicles with complex branching papillae, nuclear pseudo inclusions and nuclear grooves suggestive of papillary carcinoma of thyroglossal cyst.

facilities for frozen section biopsy were not available in our institute, we sent the entire specimen for histopathological examination (Fig. 2). Post operative recovery of the patient was uneventful.

Histopathological examination by HE stain (10 x 10 magnification) showed the cyst wall containing thyroid follicles and complex branching papillae with a central fibro vascular core lined with the cells having ground glass nuclei, nuclear pseudo inclusions and nuclear grooves. This was consistent with a diagnosis of papillary carcinoma of thyroglossal cyst (Fig. 3). On the basis of this histopathological report we performed total thyroidectomy after two months. This was followed by routine thyroxine supplementation. There was no sign of recurrence even after two years of follow up.

Case 2

A 30-yr-old female presented with midline neck swelling for 5 yrs. Initially it was small in size, but had progressed gradually, interrupted by repeated episodes of infection. On clinical examination, the mass was 0.75 x 0.5 cm in dimension, situated in midline, non tender, non pulsatile, moved on deglutition as well as on protrusion of tongue. Thyroid gland was normal on palpation and no palpable cervical lymphadenopathy could be detected. FNAC of the mass was suggestive of thyroglossal cyst. USG of thyroid gland as well as thyroid scan, done pre operatively was within normal limits as in the first case.

The operative procedure was the same as done in the first case. Postoperative histopathological examination was consistent with the diagnosis of papillary carcinoma of thyroglossal cyst. There was however no evidence of capsular invasion and margins were free of any malignant change. Repeated evaluation of thyroid gland, both clinically and radiologically, failed to validate any indication for thyroidectomy. Patient had been kept under close observation and there had been no evidence of recurrence after one year of follow up.

Discussion

Thyroglossal cysts are the most common non odontogenic midline neck swelling with a frequency of 70% in children and 7% in adults [3]. The development of thyroid gland starts by fifth week and continues till seventh week of embryogenesis. During the descent, the communication between the thyroid gland and foramen caecum, which usually disappears, may occasionally persist to form thyroglossal duct. Secretions from the cells lining the duct accumulate to form thyroglossal cyst. Malignant transformation of thyroglossal cyst is very rare and overall incidence varies from 0.7 to 1% as reported in various medical literature [2, 3].

The first case of thyroglossal cyst carcinoma was reported in 1915 and till now only a few cases have been re-

ported [1, 4, 5]. Females are more commonly affected than males with ratio being 3:2. The exact etiology of neoplastic change is not known, but de novo origin and spread from a primary thyroid gland tumour are two probable school of thoughts [6]. Radiation exposure is an important etiological factor worth mentioning.

Patients usually present with an asymptomatic midline neck mass and neoplastic changes are very difficult to apprehend beforehand. However rapid increase in size, dysphagia, hoarseness of voice and pain may be the harbingers of neoplastic change [3]. Though radiological evaluation by USG and CT scan are done regularly, the diagnosis is confirmed only by histopathology [7]. Diagnostic criteria as suggested by Joseph and Komorowski included the presence of malignant cells in the duct or cyst combined with squamous epithelial lining, normal thyroid follicle nests in the duct or cyst wall and the presence of normal thyroid gland. The most common histological types are papillary carcinoma (80%), mixed follicular pattern (8%), squamous cell carcinoma (6%), follicular carcinoma (3%), adenocarcinoma and various unclassified tumors (3%) [8]. Coexistence of two different histopathological types have also been mentioned in same cyst. Papillary carcinoma carries the best prognosis with the cure rate of more than 95 % [3, 7].

Removal of thyroglossal cyst by standard Sistrunk operation is the treatment of choice [3, 7]. Kristensen et al suggested absence of lymph node involvement and non invasion of tumour through the cyst wall as criteria for the safety of this procedure [9]. Sistrunk operation should be combined with total or sub total thyroidectomy if there is cyst wall invasion, palpable cervical lymphadenopathy or if the size of cyst carcinoma is more than 1cm. This is usually followed by radio iodine ablation and T₄ suppressive therapy [7]. The concept of total or sub total thyroidectomy as a treatment modality for thyroglossal cyst carcinoma is similar to papillary carcinoma of thyroid due to their common embryological origin and multicentric lymphatic spread. Neck dissection is performed only for proven cases of cervical lymph node metastasis [3]. USG of neck and thyroglobulin estimation may be helpful for detecting any recurrence [3, 7].

In our first case, the diagnosis of thyroglossal cyst carcinoma in the post operative biopsy report along with the size of the cyst demanded total thyroidectomy. However, in the second case, we could not justify any valid reason for

revision thyroidectomy. Absence of capsular invasion, size of the tumour and no palpable cervical lymphadenopathy justified our approach.

Squamous cell carcinoma, in contrast to papillary carcinoma carries a dismal prognosis. Sistrunk operation with post operative radiation to both sides of neck is the standard treatment protocol even in absence of regional lymphadenopathy [3].

It can be concluded hence that though thyroglossal cyst carcinoma is a rare entity, the possibility of a thyroglossal cyst which we commonly encounter in Otolaryngology practice, being a malignant one, should be kept in mind. Moreover the management of thyroglossal cyst carcinoma definitely possesses a therapeutic challenge to the surgeons. Owing to rarity of the condition, controversies do exist regarding optimal strategy to be adopted. Management of cases thus needs to be tailored individually.

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