Indian Journal of Surgical Oncology (December 2019) 10(4):676–678
https://doi.org/10.1007/s13193-019-00977-8

CASE REPORT

A Case Rarity: Papillary Thyroid Carcinoma with Squamous Metaplasia Complicated with Chronic Discharging Ulcers

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Received: 15 June 2019 / Accepted: 7 August 2019 / Published online: 2 September 2019

Abstract

We report a unique case of differentiated thyroid carcinoma (DTC) with squamous metaplasia complicated with chronic discharging ulcer. A 76-year-old gentleman was referred to us after defaulted treatment 1 year post-total thyroidectomy. He presented to us with long-standing chronic, non-healing, ulcerative and discharging wound at the anterior neck at previous total thyroidectomy scar. The primary tumour was histologically diagnosed as papillary malignant cells with extensive squamous metaplasia. Squamous metaplasia is a rare finding in thyroid carcinoma that carried a poorer prognostic factor.

Keywords Papillary thyroid carcinoma · Squamous metaplasia · Discharging ulcer · Fatal

Introduction

Although squamous metaplasia is known to be associated with papillary thyroid carcinoma [1, 2], it is an uncommon finding and this case review is about a unique case of papillary thyroid carcinoma with squamous metaplasia complicated with chronic discharging ulcers post-total thyroidectomy. Patient did not receive any radioiodine ablation therapy post-operation as he defaulted treatment. We also highlighted a few issues on devising a treatment plan for this patient when he presented to us a year after his thyroid surgery.

Case Presentation

A 76-year-old gentleman initially presented with progressive anterior neck swelling for 3 years. It was associated with lethargy, loss of appetite, dysphagia and hoarseness of voice.

Computed tomographic (CT) scan of neck showed complex right thyroid cystic nodule with local extension and mass effect to adjacent structures. After few counselling sessions, tumour debulking and total thyroidectomy were performed, where a huge thyroid tumour of the size of 14 cm × 8 cm with cystic and solid component was found adhered to the surrounding muscles. Histopathological examination revealed

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Fig. 1 Neck mass with two ulcerative wounds

papillary structures composed of papillary malignant cells with extensive squamous metaplasia.

Post-operatively, his disease was complicated with occurrence of swelling and inflammation over the wound, within first week post-surgery, which required treatment with antibiotics. Culture of seropurulent discharge showed positive for *Staphylococcus aureus*.

He was started on thyroid hormone replacement (THR) therapy with L-thyroxine for thyroid-stimulating hormone (TSH) suppression and was referred to our nuclear medicine unit for radioiodine therapy. However, he was not compliance to L-thyroxine and defaulted subsequent treatment.

A year later, he presented again with complaint of wounds over his neck swelling. On examination, there was an anterior mass, hard in consistency and fixed to the underlying structures measuring about 8 cm × 6 cm with presence of two ulcerative wounds (Fig. 1).

His thyroid function test showed low T4, TSH of 3.4 mIU/L, serum thyroglobulin level of 17.8 μg/L and anti-thyroglobulin antibody of 16.6 IU/L. Diagnostic whole body scan with 5 mCi of Iodine-131 showed uptake confined to the neck region with no evidence of metastasis elsewhere (Fig. 2). He was planned for re-biopsy of the neck mass for tissue type and to look for possibility of dedifferentiation of tumour cells. CT scan of neck was planned to look for the extension of the mass. However, his disease rapidly progress and worsen.

Discussion

This case highlights the progression of the disease post-total thyroidectomy in patient who is not compliance to the THR therapy and did not receive radioiodine treatment. It also highlights the local aggressiveness of the disease and also the potential issues in treatment plan for such a patient.

Papillary thyroid carcinoma accounts for about 80% of all thyroid carcinoma [3–5]. Generally, the prognosis of this disease is good with a survival rate of more than 95% at 25 years [4] although some factors do change the prognosis, such as age more than 45 at diagnosis [3], tumour size [4, 5], certain histopathology variants such as tall cell, columnar and diffuse sclerosing variant and presence of local invasion [6]. Squamous metaplasia is an uncommon finding in thyroid carcinoma and thought to be derived from remnant of thyroglossal duct, thymic epithelium or ultimobranchial remnants [7] or can be due to chronic inflammation or scarring from thyroiditis [7, 8]. Patients with squamous metaplasia carried a poor prognostic factor and also the risk of transformation into pure squamous cell carcinoma or dedifferentiation into anaplastic form [1, 8].

In this case, the remnant post-total thyroidectomy developed into a local mass quite rapidly within the first week after surgery, possible due to the locally aggressive process of the squamous metaplasia [9]. It was attributed to the infection as evidenced by the culture of the aspirated fluid, although *Staphylococcus aureus* can be a skin contaminant causing false positive result of the culture.

Subsequently, two chronic ulcerative discharging wounds developed at the neck with two possible reasons. Firstly, patient was not compliance to L-thyroxine causing low T4 level

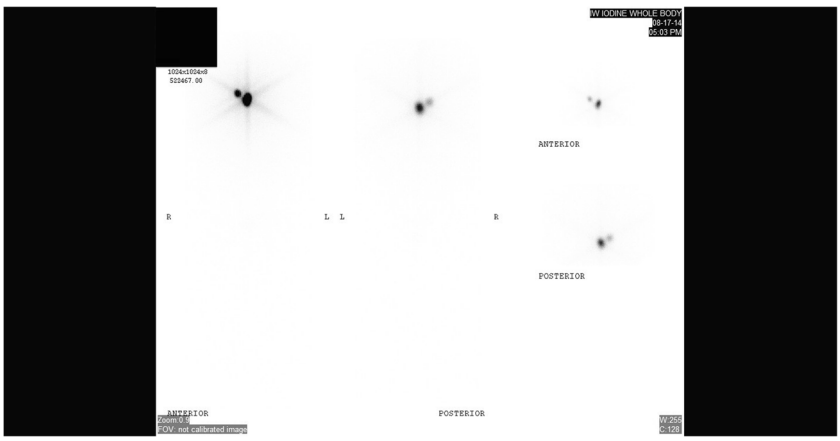

Fig. 2 Iodine-131 diagnostic whole body scan showing uptake over the neck region

or hypothyroidism, which is a known cause for delayed wound healing [10]. Secondly, it is also likely due to the local infiltration of the disease to the skin [9], possibly aggravated by the multiple needle aspiration done post-operatively to drain out the seropurulent fluid. The disease was indolent for the past 1 year even without treatment.

As the patient presented to us, there were a few issues pertaining to the treatment plan. What will be the aim of treatment for this patient: curative or palliative? This determines how aggressive the treatment plan will be. Ideal treatment plan would be combination of surgical excision of the mass followed by radioiodine ablation and external beam radiotherapy and long-term thyrotropin suppression [11]. Considering the patient's age with the background of hypertension, should we subject the patient to another round of surgery to excise the neck mass as there are risks of surgical complications of the distorted anatomy and tissue adherence intra-operatively to impaired wound healing process, wound infection and recurrent nerve palsy post-operatively [12].

There will also be problems administering Iodine-131 therapy. Firstly, with the local neck mass, administering high-dose Iodine-131 might cause neck oedema and radiation thyroiditis leading to obstructive symptoms [13] with dire consequences. Secondly, the discharging wounds pose a significant risk of contamination, both to the patient and the caregiver. Frequent dressing needs to be done for the wound with proper disposal of the waste as Iodine-131 will be accumulated in the wound discharge; thus, the patient should be kept in-patient during the course of treatment and the staffs in charge must be fully aware and protected during the changing of dressing.

Although the metaplastic components are usually well-differentiated [2], the squamous metaplasia component might not have the affinity for iodine. Thus, radioiodine ablation might not be effective and patient will need to receive radiotherapy [7]. Even after the treatment, it is pertinent that the patient be compliant to the L-thyroxine for adequate suppression of TSH [11] while maintaining a normal T4 level. Compliance will be an issue as patient has history of defaulting his treatment. Thus, patient and family members will need to be properly counselled and all factors that might lead to default of treatment will need to be explored and discussed.

In conclusion, this case review highlights the occurrence of a rare case of papillary thyroid carcinoma with squamous metaplasia complicated with chronic discharging ulcers post-total thyroidectomy where patient defaulted radioiodine ablation therapy for a year and the potential issues in treatment planning for such a patient which clinicians should be aware of.

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