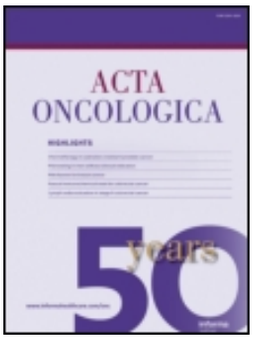

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LETTER TO THE EDITOR

Papillary carcinoma in a thyroglossal duct

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To the Editor

The thyroid gland starts its development in the fourth week of gestation from a segment of endodermal cells in the floor of the primitive pharynx, subsequently descending in the neck as part of the thyroglossal duct, and reaching its endpoint at the thyroid cartilage by the end of the seventh week. It consists of cuboidal or columnar epithelium and normally involutes after the descent of the thyroid gland has been completed. However, in approximately 7% of the population, remnants of the thyroglossal duct persist and may lead to the formation of a thyroglossal duct cyst.

Adequate treatment of a benign thyroglossal duct cyst consists of complete resection of the epithelium-walled ductal remnants. However, simple cyst excision results in a high recurrence rate of up to 30–60% [1,2], probably due to unidentified ductal remnants in the cranial part of the duct. Radical excision of the cyst and thyroglossal duct tract including excision of the midportion of the hyoid bone and tract excision to the foramen coecum as proposed by Sistrunk [3] results in a lower recurrence rate of approximately 3% [1,4]. A significant proportion (5 to 45%) of thyroglossal duct cysts contains normal ectopic thyroid gland tissue [5,6]. Malignant degeneration is rare.

Recently, a 30 year old man presented to us with a 3-month history of a slowly growing mass in the midline of the neck. There was no tenderness or drainage. Physical examination revealed an approximately 5×2 cm soft mass with some extension to the left side of the neck. No associated cervical lymphadenopathy was present. Ultrasound showed a round cystic fluid collection without signs of enlarged cervical lymph nodes. Fine needle aspiration revealed clear fluid with some macrofages and granulocytes, but no signs of malignancy.

Subsequently, a Sistrunk procedure was carried out, removing a $5 \times 2 \times 1$ cm thyroglossal duct cyst including excision of the median one third of the hyoid bone and a core of tissue up to 1 cm of the supra-hyoid portion of the tract. Postoperatively, recovery was uneventful.

Pathologic examination of the specimen showed an irregular mass of $4.5 \times 1.5 \times 1$ cm with a small portion of bone attached. Cut surfaces revealed a large cyst with a smooth wall and filled with haemorraghic material. Some mucus formation was present. Macroscopically, the lesion was excised in total.

Microscopic sections revealed a partially cystic lesion with haemorrhagic intra-luminal contents and fibrosis, sclerosis and infiltration in the wall. Ectopic thyroid follicular structures were focally present within the cyst wall. However, a localisation of a papillary thyroid carcinoma was identified, partially with a more irregular follicular pattern. The papillary structure was covered by cylindrical epithelium and seemed to grow closely to the muscle and the periost. No pathologic lymph nodes were found.

Postoperative imaging of the thyroid gland using ultrasound did not show any abnormalities. Considering the size of the tumor and the intimate relationship with surrounding structures, extension of the tumor through the cyst wall was suggested, and a total thyreoidectomy was deemed necessary.

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During this operation, no pathologic lymph nodes were identified.

Microscopic examination of the specimen obtained during this second operation revealed normal thyroid tissue without clues of malignancy. Subsequent, a radioactive Iodine whole body scan was performed after administration of 39 MBq iodine 131, revealing limited Iodine uptake in a lesion median in the neck. No pathologic Iodine uptake outside the original thyroid resection site was seen. The patient was treated with a 2.0 GBq iodine 131 ablation dose, followed by levothyroxine suppletion, leading to TSH suppression.

Thyroglossal duct carcinoma is a rare entity, developing in less then one percent of patients with a thyroglossal duct cyst. Sporadic cases have been reported in pediatric patients [7] and adults around the world [8–12]. The most frequent malignancy is papillary adenocarcinoma (80%), followed by mixed papillary and folliculary carcinoma (7%), follicular carcinoma (3%), squamous cell carcinoma (5%) and various unclassified malignancies (5%) [5,13]. Medullary thyroid carcinoma has never been reported in a thyroglossal duct cyst. The existence of thyroglossal duct carcinoma has been disputed. Some authors stated papillary thyroglossal duct carcinoma should be considered a local metastasis of a thyroid tumor, the thyroglossal duct serving as a natural conduit for tumor cells [14,16]. However, ectopic thyroid follicles are found in 5% to 45% of thyroglosal duct cysts, depending how thoroughly the tissue surrounding the cyst has been examined [6]. Joseph [5] proposed the following criteria for the unequivocal diagnosis of primary thyroglossal duct carcinoma: 1) finding of a thyroglossal remnant that can be distinguished from a cystic lymph node metastasis by a columnar or squamous epithelial lining, 2) presence of a few normal thyroid follicle nests on the cyst wall and 3) presence of a normal thyroid gland. However, Mobini controversially stated squamous cell carcinoma is probably the only true carcinoma of the thyroglossal duct, since the other malignancies develop in ectopic thyroid tissue [15]. Squamous cell thyroglossal cyst carcinoma is extremely rare, and probably arises from metaplastic change from columnar to squamous epithelium in the cyst wall due to high pressure.

In the majority of cases, the diagnosis of thyroglossal duct carcinoma is made postoperatively after microscopic review of what was presumed to be a benign thyroglossal duct cyst [16]. However, malignancy must be suspected in all cases of thyroglossal duct cyst with recent changes in clinical behaviour or after previous local radiation. Preoperative ultrasound imaging or computer tomography does not allow a certain diagnosis, although asymmetrical thickening of the cyst wall warrants further imaging. Fine needle aspiration yields a correct diagnosis in only 66% of the cases.

The most advocated surgical treatment for thyroglossal duct carcinoma is no additional treatment to the Sistrunk procedure [13,16–18] if the criteria proposed by Kristensen [16] are fulfilled: 1) histologically normal ectopic thyroid follicles are found in the cyst; 2) the tumor has not extended through the cyst wall; 3) the thyroid gland is normal; and 4) no cervical lymph node involvement is present.

Other authors [19] advise total thyreoidectomy and subsequent iodine 131 ablation dose to eliminate the 10–30% risk of a second focus of papillary or follicular adenocarcinoma in the thyroid gland.

In case of large tumors (>1 cm), invasion through the duct cyst wall or suspect foci in the thyroid gland, a thyreoidectomy followed by iodine 131 ablation and TSH suppression is the most frequently proposed treatment[13,16–18]. In cases of midline cystic lymph node metastasis of thyroid carcinoma mimicking thyroglossal duct carcinoma, a total thyreoidectomy with subsequent iodine 131 ablation dose is strongly advised. Discussion still exists if cervical lymph node metastasis should be treated by bilateral radical neck dissection or selective lymph node picking [20].

Multifocal growth is a well-known problem in papillary thyroglossal duct carcinoma. In approximately 10% a second malignant focus will become present in the thyroid gland [21]. Moreover, papillary carcinoma arising from ectopic thyroid tissue in a thyroglossal duct has the capacity of metastasizing to regional lymph nodes [18]. This warrants rigid follow-up, both clinically and radiologically [9,21].

The prognosis for papillary thyroglossal duct carcinoma is probably similar to, or even better than that of tumors in the thyroid gland, with cure rates up to 95%. Squamous cell thyroglossal duct carcinoma has a considerably worse prognosis with at least 30–40% mortality in small series of this rare tumor [11,17]. This prognosis appears to be slightly more favourable than squamous cell carcinoma of the thyroid gland, which carries an infaust prognosis in most patients within one year. Complete local excision is imperative because recurrence rate is high.

We conclude papillary carcinoma arising in a thyroglossal duct cyst is a rare entity. This malignant tumor can be well treated performing a Sistrunk procedure if Kristensen's criteria are fulfilled. If these criteria are not fulfilled (as was the case in our patient), we strongly suggest total thyreoidectomy with subsequent Iodine 131 ablation, leading to a good to excellent prognosis.

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