MANAGEMENT OF MALIGNANT THYROGLOSSAL DUCT CYSTS, A RARE CLINICAL ENTITY IN CERVICAL TUMOR PATHOLOGY

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MANAGEMENT OF MALIGNANT THYROGLOSSAL DUCT CYSTS, A RARE CLINICAL ENTITY IN CERVICAL TUMOR PATHOLOGY (Abstract): Thyroglossal duct cyst is a congenital malformation that occurs at cervical level following an embryonic developmental defect. The authors undertook a retrospective study on 143 patients diagnosed with malignant thyroglossal duct cyst in the ENT Clinic of “Sf. Spiridon” County Clinical Emergency Hospital Iasi, between 1992 and 2016 and only 2 cases came out as malignant thyroid gland cyst. This confirms the rarity of this type of lesion, consistent with the data presented in the literature. We present the case of a 51-year-old male case, hospitalized in the ENT Clinic of “Sf. Spiridon” County Clinical Emergency Hospital Iasi, treated by surgical excision of the thyroid gland cyst, performed by using the Sistrunk method. The post-operative histopathological examination of the thyroid gland cyst detected the malignancy. Conclusions: The oncology committee of the hospital decided to complete the therapeutic protocol by total thyroidectomy, bilateral neck dissection, radioiodine administration, followed by thyroid hormone replacement. Regular follow-up showed a favorable outcome without complications and no tumoral recurrence at 5 years. Keywords: THYROGLOSSAL MALIGNANT DUCT CYST, HISTOPATHOLOGY, RADIOLOGY, SURGERY, ONCOLOGICAL TREATMENT.

INTRODUCTION
Thyroglossal duct cyst is a congenital malformation that occurs at cervical level following an embryonic developmental defect. The thyroid gland developed at the foramen cecum level migrates to the lower third cervical level. The existing tract normally closes after birth. The persistence of this tract can generate the thyroid gland cyst, located between the base of the tongue and the thyroid, with an internal structure of respiratory epithelium and possible thyroid follicles at the external wall of the cyst. The formation is located in the antero-medial cervical section, below the hyoid bone and more rarely the left. It is initially asymptomatic and becomes manifest after an inflammatory episode (1, 2, 3).

In evolution, a fistula can develop, most frequently externally and rarely internally in the pharynx, due to infection. Rarely, the thyroid follicles can undergo malignant transformation. This malignant degeneration most commonly occurs as a thyroid papillary carcinoma developed from the cyst wall.

Differential diagnosis should exclude ectopic thyroid, adenopathy, dermoid cyst, Schwannoma and cystic lymphangiomas (4, 5). In case of thyroid gland cyst malignan-
cy, the accepted therapeutic protocol, that is also cited in the literature, is the complete excision of the thyroid gland and cervical lymph nodes, with post-surgical radiiodine administration followed by thyroid hormone replacement (6, 7).

Between 1992 and 2016, in the ENT Department of “Sf. Spiridon” Hospital from Iasi, Romania, 143 patients were diagnosed with thyroglossal duct cysts, 15 developed thyroglossal duct fistulas, out of which 2 cases were histopathological diagnosed as thyroglossal malignant duct cyst.

Thyroglossal duct cysts and fistulae predominates in the third and fourth decade of life (93 patients), respective 45 patients were female, and 98 patients were male.

The positive diagnosis was based on physical examination combined with cervical echography, scintigraphy, histopathological exam and CT scan for some cases. Patients with thyroglossal duct cysts had a prior asymptomatic mobile neck mass, which gradually expanded, located in the midline or left lateral part of the neck, usually under the hyoid bone. Some thyroglossal duct cysts develop infectious processes and a cutaneous fistula might appear.

All surgeries were performed under general anesthesia. Because the embryological development of the hyoid bone is closely linked to thyroglossal duct cysts, the middle portion of the hyoid bone was excised with the cyst (Sistrunk procedure). In case of malignant cysts, the protocol of the oncologic committee was applied. In five patients who presented fistulae, methylene blue was injected to ease the dissection of the thyroglossal duct cysts.

CASE REPORT

We present the case of a 51-year-old male case hospitalized in the ENT Clinic of “Sf. Spiridon” County Clinical Emergency Hospital Iasi for a cervical mass located below the hyoid bone, which occurred and was observed 2 months ago before being hospitalized, after one episode of upper respiratory tract viral infection. The ENT clinical examination revealed an elastic mass under the hyoid bone, mobile with swallowing.

Cervical echography associated with an endocrinological examination detected a well-defined cystic mass under the hyoid bone stretching to the anterior border of the thyroid cartilage with a normal appearance and topography of the thyroid gland (fig.1).

![Fig. 1. Echographic image of the cervical mass](image-url)
The dosage of thyroid hormones was within normal range.

The elected treatment was surgery under general anesthesia by external cervical approach. We performed the dissection of the mass which was adherent to the external perichondrium of the thyroid cartilage, the cranial part being adherent to the hyoid bone, having a tract to the base of the tongue. The whole cyst together with the body of the hyoid bone was excised, ligating the duct above the level of the hyoid bone, using the Sistrunk method (fig. 2-5). The histopathological result showed a cyst wall made up of fibrous tissue with moderate chronic inflammatory microhemorrhage infiltration and a cholesterol deposit, somewhere tapped by a squamous epithelium. Aspects of thyroid papillary carcinoma developed on the thyroid follicles where identified in the wall of the cyst. The examined sections identify adipose tissue and striated muscles with no tumor infiltration and bone fragments without tumor infiltration. The histopathological aspect corresponds to a thyroid papillary carcinoma, pT2NxG2 (fig. 6).

Immunohistochemical (IHC): CK19 - positive in malignant tumor cells, HBME-1 - positive in malignant tumor cells.

The histopathological aspects identified and the IHC profile corresponds to a thyroid papillary carcinoma originating in a thyroid gland cyst.

**Fig. 2.** Incision revealing the cyst  
**Fig. 3.** Dissection of the cyst  
**Fig. 4.** Post excision appearance  
**Fig. 5.** Excised specimen (cyst + hyoid bone)
Due to the malignant transformation of the thyroid gland cyst, it required the completion of the therapeutic protocol by performing second surgery for excision of the thyroid gland (total thyroidectomy), followed by application of radiotherapy according to a protocol conducted by the oncological committee.

The postoperative outcome was favorable, with follow-up performed at 1 month, 3 months, 6 months, one year and then annually up to 5 years, without tumor recurrence but requiring thyroid hormone replacement.

DISCUSSION
Thyroid gland cyst, frequent in young adults, is most commonly manifested by a respiratory infection that causes cyst superinfection with increased volume and possible appearance of a fistula.

The need for differential diagnosis to exclude other cervical swelling and thyroid gland pathology is imperative. For this purpose, radiological paraclinical examinations are performed and the patient is endocrinologically examined preoperatively (2).

Endocrinological evaluation, as well as the dynamic outcomes of hormone replacement by administration of Eutirox showed an optimal hormonal substitution level with clinical development in normal parameters that allowed normal social reinsertion of the patient.

The particularity of this case was the malignant transformation of the thyroid gland cyst, the second case encountered in our ENT department over the last 25 years, which corresponds to other statistics stated in the medical literature (7, 8).

A multidisciplinary team consisting of ENT surgeons, radiologists, endocrinologists, histopathologists and oncologists was needed in order to offer the best treatment plan for the patient (9, 10, 11).

The rest of the non-malignant thyroglossal duct cyst cases were resolved by using the Sistrunk surgical procedure, without postoperative complications and no relapse of the tumor (12).

CONCLUSIONS
Malignant thyroglossal duct cyst represents a rare complication of a congenital malformation in ENT pathology, diagnosed postoperatively following the histopathological examination.

The oncological therapeutic protocol requires excision of the thyroid gland as a complementary step, if considering the phylogenetic development of thyroid gland and thyroid gland cyst, followed by radioiodine administration and hormone replacement.

The surgical approach by block excision of the cyst and the hyoid bone body, with the ligature at the base of the tongue (Sistrunk procedure), prevents local relapse and prevents loco-regional complications.

The social reinsertion, the prognosis and the evolution of the patient under the conditions of applying a correct oncological protocol was favorable.
REFERENCES


NEWS

THE ROLE OF MATERNAL OMEGA-3 FATTY ACIDS IN METABOLIC DYSFUNCTIONS IN OFFSPRING

A study by Robertson et al. analyses the role of maternal diet during gestation and lactation on the development of the offspring’s intestinal microbiota. Metabolic dysfunctions in adult life are influenced by the development of the early-life intestinal flora, which, in turn, is dependent on maternal fatty acid status. To prove this, Robertson et al used a transgenic model and showed that maternal n-3 polyunsaturated fatty acid production during gestation or lactation has different effects on male offspring compared to female offspring. Thus, maternal production of polyunsaturated fatty acid significantly reduced weight gain in males fed a high-fat diet but had no significant effect in female offspring. This effect on males persisted into adulthood life and was mediated by restructuring of gut microbiota composition. The microbiota in offspring is influenced more by the maternal fatty acid profile during lactation than in utero. The study shows that reduced exposure to maternal n-3 polyunsaturated fatty acid leads to significantly depleted Bacteroides, Akkermansia and Epsilonproteobacteria and higher relative abundance of Clostridia (Robertson RC, Kaliannan K, Strain CR, Ross RP, Stanton C, Kang JX. Maternal omega-3 fatty acids regulate offspring obesity through persistent modulation of gut microbiota. Microbiome 2018; 6(1): 95. Published 2018 May 24. doi:10.1186/s40168-018-0476-6).