HURTHLE CELL CARCINOMA IN CHILDHOOD: RETROSPECTIVE ANALYSIS OF A LARGE SERIES

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BACKGROUND

In general population Hurthle cell cancer (HCC) accounts for 3-7% of all differentiated thyroid cancers (TC) with a more aggressive course, while its relative prevalence and behavior in childhood is uncertain due to the lack of specific literature reviews.

OBJECTIVES

To describe the largest pediatric HCC cohort to date reported and to estimate its relative prevalence among TC variants in childhood.

METHODS

Study population included 5 patients <19 years, who were diagnosed with HCC during the period 2000-2018 in our Departments. Histologic diagnosis of HCC was based on the finding of at least 75% of Hurthle cells at post-surgical analysis.

HCC course was retrospectively reconstructed with data recorded at diagnosis, at surgical resection and during follow-up. Patients' assessment included: clinical findings, thyroid function and autoimmunity tests, neck and chest imaging (ultrasound and computed tomography scan), cytologic and histologic analyses of the tumor.

Table 1

Patients (n)	1	2	3	4	5
Sex	M	F	F	M	F
Age (yrs)	7.1	12.3	12.5	14	16.7
Biochemical picture					
Euthyroidism	+	+	+	+	+
Maximum nodule diameter (cm)	3.7	3.5	2.0	1.3	2.0
Cytologic stages [a]	III	IV	III	III	IV
Presence of Hurthle cells at cytology	+	-	-	+	-
Presence of Hurthle cells at histology (%)	>75	100	100	100	>75
Risk levels [b]					
Low	+		+	+	
Intermediate		+			+
Pathological antecedents:					
Hashimoto's thyroiditis					+
Neurofibromatosis Type 1	+				
Osteosarcoma			+		
Follow-up duration (yrs) [c]	6.5	1.5	8.9	15.1	14.3

RESULTS

HCC occurred with a relative prevalence of 5.8% (5 of 86 young patients affected by TC, diagnosed in the same period and institutions), at a median chronological age of 12.5 years.
All patients were biochemically euthyroid at HCC diagnosis and underwent both total thyroidectomy with central neck dissection and radioiodine therapy, with subsequent L-T4 thyroidal suppression.
Low or intermediate risk level was observed at diagnosis, since none of our patients exhibited extensive lateral neck disease or distant metastases and all of them showed a persistent clinical, biochemical and imaging remission over time.
Antecedents of other diseases were recorded in 3 patients (Hashimoto's thyroiditis, Neurofibromatosis type 1 and osteosarcoma respectively).

Table 1. Clinical, biochemical, cytologic and histologic features and risk levels in the 5 children studied at the time of diagnosis and post-therapy follow-up duration.
[a] According to the Bethesda system (1)
[b] According to the staging system of the American Thyroid Association task force for children with thyroid nodules and differentiated cancer, described by Francis et al. (2)
[c] From the onset of treatment to the last examination

CONCLUSIONS

- In childhood the relative prevalence of HCC among TC histotypes is **5.8%**, that is close to the one reported from literature both in general population (3-5.7%) and in young patients (2-7%);
- HCC may develop **even very early**, at an age of 7 years;
- in pediatric age HCC does not seem to have a more aggressive behavior than other TC histotypes;
- antecedents of other diseases are not infrequent in the history of children with HCC.

References:

1. Cibas ES & Ali SZ. The 2017 Bethesda System for Reporting Thyroid Cytopathology. *Thyroid* 2017 27 1341-1346.
2. Francis GL, Waguespack SG, Bauer AJ, Angelos P, Benvenga S, Cerutti JM, Dinauer CA, Hamilton J, Hay ID, Luster M, et al.; American Thyroid Association Guidelines Task Force Management Guidelines for Children with Thyroid Nodules and Differentiated Thyroid Cancer. *Thyroid* 2015 25 716-759.