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Review

Primary thyroid angiosarcoma: A systematic review

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ABSTRACT

Thyroid angiosarcoma (TAS) is rare and represents a very aggressive malignancy. Its rarity is principally linked to two major pitfalls. Firstly, TAS histopathology diagnosis can be difficult; second, the limited clinical experience with this condition can make its management complex.

We conducted a detailed systematic review, focusing on the knowledge available regarding TAS etiopathogenesis, treatment options and prognosis. The aim is to present the main TAS characteristics and to summarize the clinical experiences described worldwide, in order to provide a useful clinical tool.

Introduction

Thyroid angiosarcoma (TAS) represents a rare malignancy and constitutes less than 1% of all sarcomas [1]. TAS is an aggressive disease, characterized by severe local course and rapid metastatic dissemination, leading to a poor prognosis. It arises from endothelial cells. In the past years, the real existence of TAS as primary tumor in the thyroid gland has been debated due to its unusual pathological finding [2]. The distribution is predominant in female and primarily affects elderly patients with a history of goiter and rapidly enlarging neck masses [1].

Literature on this topic is scarce and has never been compiled to provide a comprehensive overview of the management of these patients. The purpose of this article is to perform a systematic review of the literature, in order to discuss the main characteristics – biologic and immunohistochemical findings, demographics and clinical course, treatment options and prognosis – and to provide some directions for future research. The aim is certainly to be of help by sharing this rare tumor within reference daily clinical practice.

Methods and materials

The preferred reporting items for systematic reviews and meta-analyses (PRISMA) statement was followed. A literature search using Pubmed databases was performed up to January 2018 without any restrictions on publication date to identify all published studies that evaluated TAS. PubMed search was performed using the following combinations of research criteria: "thyroid", "angiosarcoma", "sarcoma", "epithelioid", "hemangioendothelioma" and "fine needle aspiration". Two independent reviewers selected the identified studies based on the title and abstract; in cases where the study topic could not be clearly ascertained from the title or abstract, the full-text version was retrieved for evaluation. The search was restricted to English-language papers. Only articles with full texts were included in the analysis. Reference lists of previously published reviews were explored. When two articles appeared to report results with overlapping data, only the data representing the most recent publication or with the larger sample size were included in the analysis. Although every attempt was made to eliminate redundancy in the data represented in our meta-analysis, we cannot rule out the possibility that a few individuals participated in more than one study. Data from each study were tabulated and included first author' surname, year of publication, sample size, histological and immunohistochemical features, treatment details, clinical outcomes and duration of follow-up.

Results

The literature search identified a total of 58 potentially relevant articles on TAS. Twenty-six articles were excluded because they were not written in the English language (n = 24) or full texts were not available (n = 2). The reviewed article types included case reports and retrospective case series; no randomized trials were found. A total of 32 retrospective studies representing 61 patients were considered [3–34]. Details are listed in Table 1. Most reports were from Italy, only five

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Table 1
Retrospective studies for thyroid angiosarcoma.

Author	Year	Country	Patient (n)	Treatment	Histological features	Survival
Altinay [22]	2014	Turkey	1	Bilateral thyroidectomy	CD31 +, Weibel-Palade bodies+, vimentin +	15 months
Bayir [23]	2016	Turkey	1	Total thyroidectomy	CD31 +, CD34 +, vimentin +, tyroglobulin −, Ki67+, TTF1 −	7 days
Collini [24]	2016	Italy	6	Complete surgical resection (6); neoadjuvant CHT (1); adjuvant CHT (3); adjuvant CRT (1)	CD31 +, CD34 −, Ki67 +, FLI1 +, ERG +, tyroglobulin −, TTF1 −	DOD (2): 36 and 9 months; NED (3): 82, 70, 59 months; NA (1)
Couto [25]	2014	Portugal	1	Total thyroidectomy; adjuvant RT	CD31 +, CD34 −, tyroglobulin −	4 years
Kalitova [26]	2009	Czech Republic	1	Total thyroidectomy	Factor VIII-RA +, CD31 +, CD34 +, FLI1 +	2 months
Kaur [27]	2013	Baltimore	1	Total thyroidectomy	Factor VIII-RA +, CD31 +, CD34 +, tyroglobulin −	2 weeks
Lepe [28]	2017	USA	1	RT	CD31 +, CD34 +	After treatment
Yoon Moon [29]	2016	Republic of Korea	1	Complete surgical resection	CD31 +, CD34 +, TTF1 −	2 years
Prater [30]	2014	USA	1	Left hemithyroidectomy	CD31 +	NA
Rotellini [31]	2015	Italy	1	Surgical biopsy	CD31 +, factor VIII-RA +, vimentin +, tyroglobulin −	4 months after biopsy
Wiedermann [32]	2016	USA	1	Left and partial right thyroidectomy with left modified neck dissection (levels II–IV)	CD31 +	NA
Ryska [3]	2004	Czech Republic	6	N/A	CD31 +, Weibel-Palade bodies+, vimentin +, tyroglobulin −	DOD (2): 0.5 and 3 months; NED (1): 21 months; DUC (1): 24 months; NA (2)
Petronella [4]	2012	Italy	1	Total thyroidectomy	CD31 +, CD34 +, factor VIII-RA +	3 months
Cutlan [5]	2000	USA	1	Surgical resection; adjuvant RT	CD31 +, CD34 +, factor VIII-RA +, tyroglobulin −	3 years
Chan [6]	1986	China	1	Subtotal resection of the left thyroid lobe; adjuvant dadioiodine ablation	Factor VIII-RA +, Weibel-Palade bodies +	6 months
Goh [7]	2003	Singapore	2	N/A	CD31 +, factor VIII-RA +	5 months
Astl [8]	2000	Ungary	1	Neo-adjuvant RT; partial thyroidectomy	N/A	N/A
Lamovec [9]	1994	Slovenia	2	N/A	Factor VIII-RA +	N/A
Chesky [33]	1953	USA	1	Surgical resection	N/A	1 year
Maiorana [10]	1996	Italy	7	Total thyroidectomy	CD31 +, factor VIII-RA +	DOD (4): median 5 months; NED (3): 27, 32 and 66 months
Yilmazlar [11]	2005	Turkey	1	Total thyroidectomy; adjuvant CHT	CD34 +, factor VIII-RA +	12 weeks
Hassan [12]	2005	Germany	1	Total thyroidectomy	CD31 +, CD34 +, factor VIII-RA +	13 months
Rhomberg [13]	2004	Austria	12	Surgical resection; adjuvant RT (2), adjuvant CRT (6)	Factor VIII-RA +, tyroglobulin −	Median: 14 months (range, 0.5–196 months)
Lin [14]	2001	Brazil	1	None	CD31 +, factor VIII-RA +, vimentin +	N/A
Tanda [15]	1988	Italy	1	Total thyroidectomy; adjuvant RT	Factor VIII-RA +, Weibel-Palade bodies +	10 months
Isa [16]	2009	Malaysia	1	Left hemithyroidectomy; adjuvant RT	CD31 +, tyroglobulin −	7 months
Del Rio [17]	2007	Italy	1	Total thyroidectomy	CD31 +, CD34 +, factor VIII-RA +	N/A
Zouaidia [18]	2010	Morocco	1	Subtotal thyroidectomy; adjuvant RT	CD31 +, CD34 +, factor VIII-RA +, tyroglobulin −	5 months
Fulciniti [19]	2008	Italy	1	Total thyroidectomy	CD31 +, CD34 +, tyroglobulin −	ED: 15 months
Binesh [20]	2011	Iran	1	Neo-adjuvant CHT; RT	N/A	4 months
Beer [34]	2007	United Kingdom	1	None	Factor VIII-RA +	2 weeks
Proces [21]	1998	Belgium	1	Right thyroidectomy	Factor VIII-RA +, vimentin +	ED: 9 months

RT: radiotherapy; CRT: chemoradiotherapy; CHT: chemotherapy; DOD: died of disease; DUC: died of unrelated causes; NED: no evidence of disease; ED: evidence of disease; N/A: not available; TTF: thyroid transcription factor; PR: partial response.