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Head and Neck Sarcomas: Histopathological Characteristics in a Moroccan Institution

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Abstract: Head and neck sarcomas are exceedingly rare, representing 4–10 per cent of all sarcomas and less than 1 per cent of head and neck malignanties. The objective of this study is to analyse epidemiological and histopathological data of these neoplasms in Fez, Morocco. This is a retrospective study including all head and neck sarcomas histologically proven between 2007 and 2016. It was conducted at the department of pathology of the Hassan II university hospital, fez, Morocco. The characteristics of patients including age, sex, location, and histological diagnosis were analyzed. 54 cases of head and neck sarcomas were diagnosed. There was a male predominance with a sex-ratio of 1.5. The mean age was 37.7 years (1 year-88years). There were 45 soft tissue sarcomas, mostly located in the face, and 9 bone sarcomas mostly located in the mandible. Rhabdomyosarcoma and leiomyosarcoma were the commonest types. In Fez, head and neck sarcomas share some characteristics with previous published series and present some differences. They occur mostly in males, with a younger age at diagnosis and a higher proportion of children. Soft tissue location is the most frequent, particularly facial and rhabdomyosarcoma and leiomyosarcoma are the commonest types.

Keywords: Sarcoma; head and neck; bone; soft tissue; histopathology

INTRODUCTION

Sarcomas are a group of malignant neoplasms of mesenchymal origin that comprise less than 1% of all cancers [1, 2]. They demonstrate aggressive biological behaviour, with the majority being locally invasive with significant potential for metastasis [1]. Head and neck sarcomas are exceedingly rare, representing 4–10 per cent of all sarcomas and less than 1 per cent of head and neck malignanties [3-5]. They are less rare in pediatric population where they account for third of all sarcomas [6].

The majority of head and neck sarcomas arise from soft tissues (80 per cent) [5-7]. Bone and cartilage sarcomas represent around 20 per cent of head and neck sarcomas and less than 0.2 per cent of malignant head and neck tumours [5, 7]. In spite of the rarity of this class of neoplasms, the biologic diversity within this small number of tumors is great [8]. In general, they are grouped by mesenchymal cell of origin, head and neck subsite and histologic grade, which is a consistent predictor of prognosis [7]. Osteosarcomas, rhabdomyosarcomas, malignant fibrous histiocytomas (MFH), fibrosarcomas and angiosarcomas are the most common histologic types found in the head and neck accounting for approximately 50% of all sarcomas in this region [9]. The increased use of the immunohistochemistry and molecular oncology markers has furthered our ability to definitively subclassify sarcomas; however, 20% will still remain unclassified, highlighting the challenges that remain [10].

Numerous studies have been carried out in various countries about the epidemiological characteristics of head and neck sarcomas and numerous studies have demonstrated geographical variation of incidence, age and gender prevalence, location and histopathological characteristics. In Morocco, few papers have been published [2] and there is no available data about these tumors in the region of Fez. Therefore, we conducted this retrospective study with the aim to analyze demographical and histopathological data of these neoplasms in Fez, between 2007 and 2016.

MATERIALS AND METHODS

This is a retrospective study including all head and neck sarcomas histologically proven between 2007 and 2016. It was conducted at the department of pathology of the Hassan II university hospital, fez, Morocco. The characteristics of patients including age, sex, location, and histological diagnosis were analyzed. Age was expressed as mean and range and was divided into two groups: children (age≤18 years) and adults (age>18 years), and into various groups by decade. Sex was expressed by percentage and sex ratio. In regard to the location of tumor, it was expressed by frequency and was divided into soft tissue and hard tissue. Soft tissue was divided into the following sites: face, scalp, sinonasal cavity, orbit, larynx, nasopharynx, parotid, oral cavity, ear, neck, and thyroid. Hard tissue was divided into mandible, maxilla, ethmoid bone and larynx. Distribution of the different histological types was analysed according to age and sex.

Statistical analysis was carried out using Epi-Info7 version 7.1.0.6.

RESULTS

Age and gender

During a 10-year period, 54 cases of head and neck sarcomas were diagnosed. There were 33 males and 21 females, with a respective proportion of 61.1% and 38.9% and a sex-ratio of 1.5.

The mean age at the time of diagnosis was 37.7 years (1 year-88years). There were 13 children (24.1%) and 41adults (75.9%).

Age distribution by decades showed a peak at the third decade. Details are provided in figure1.

Location

Regarding anatomic distribution of the tumors, face, scalp, orbit and sinonasal cavity were the most frequent locations. Repartition into soft tissue sarcomas and bone and cartilage sarcomas showed 45 cases located in soft tissue (83.3%), mostly in face, and 9 located in hard tissue (16.7%), mostly in the mandible. Table 1 shows details of topographic repartition.

Histological types

Concerning histological characteristics, rhabdomyosarcoma and leiomyosarcoma were the commonest types overall. The two most frequent sarcomas in adults were leiomyosarcoma and carcinosarcoma, and in children they were rhabdomyosarcoma and leiomyosarcoma.

Analysis of sex distribution according to histological type showed that leiomyosarcoma and carcinosarcoma were more frequent in males whereas chondrosarcoma and dermatofibrosarcoma protuberans were more frequent in females. Rhabdomyosarcoma, Kaposi sarcoma and osteosarcoma were equally seen in both sexes.

Analysis of topographic distribution for each type showed that rhabdomyosarcoma was mostly located in nasopharynx, sinonasal cavity, orbit and scalp. Leiomyosarcoma was mostly located in sinonasal cavity, face and larynx. Among bone sarcomas, chondrosarcoma was frequently maxillar and osteosarcoma was frequently mandibular. Histological type distribution as well as demographic and topographic characteristics of each type is illustrated in table 2.

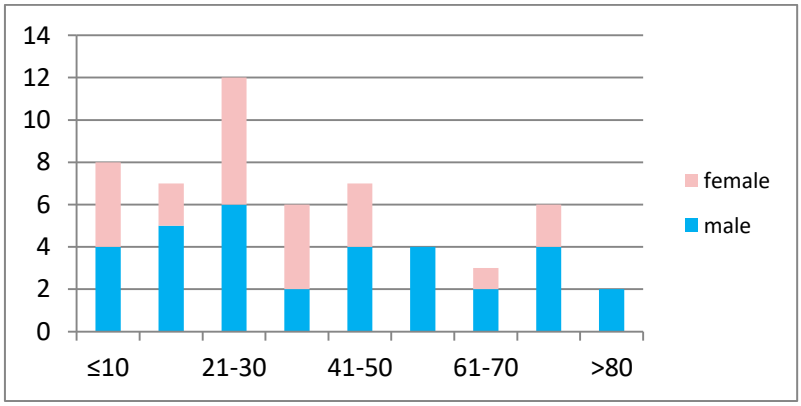

Fig-1 : Repartition according to age and gender

Table 1: Repartition according to location

	n	%
Bone and cartilage	9	16.7
- mandible	- 4	- 7.4
- maxilla	- 3	- 5.5
- ethmoid	-1	- 1.9
-larynx	-1	-1.9
Soft tissus	45	83.3
- face	- 12	- 22.2
- scalp	- 8	- 14.8
- sinonasal cavity	- 6	- 11.1
- Orbit	-7	- 12.9
-larynx	-3	- 5.5
- nasopharynx	- 3	- 5.5
-parotid	-2	- 3.7
- oral cavity	- 1	- 1.9
-ear	-1	- 1.9
-Neck	-1	- 1.9
- thyroid	-1	- 1.9
Total	**54**	**100**

Table 2: Histological type distribution and demographic and topographic characteristics of each type

	RMS	LMS	CS	KS	CHS	OSS	DFSP	MSFT	MPNST	EWS	MFH	UDS	Total
Age n													
-Adults	6	7	7	5	4	4	3	2	0	0	1	1	41
-Children	9	2	0	0	0	0	0	0	1	1	0	0	13
Sex n													
-Male	8	8	7	3	1	2	0	1	0	1	1	0	33
-Female	7	1	0	2	3	2	3	1	1	0	0	1	21
Location n													
-NPH	3	0	0	0	0	0	0	0	0	0	0	0	3
-Scalp	3	1	1	0	0	0	2	0	0	1	0	0	8
-SNC	3	2	0	0	0	0	0	1	0	0	0	0	6
-Orbit	3	1	2	0	0	0	0	1	0	0	0	0	7
-Face	2	2	2	4	0	0	1	0	0	0	1	0	12
-Parotid	1	0	1	0	0	0	0	0	0	0	0	0	2
-larynx	0	2	1	0	1	0	0	0	0	0	0	0	4
-ear	0	1	0	0	0	0	0	0	0	0	0	0	1
-thyroid	0	1	0	0	0	0	0	0	0	0	0	0	1
-OC	0	0	0	1	0	0	0	0	0	0	0	0	1
-Neck	0	0	0	0	0	0	0	0	1	0	0	0	1
-Mand	0	0	0	0	1	3	0	0	0	0	0	0	4
-Max	0	0	0	0	2	0	0	0	0	0	0	1	3
-ETH B	0	0	0	0	0	1	0	0	0	0	0	0	1
Total n (%)	**15 (27.7)**	**10 (18.5)**	**7 (13)**	**5 (9.2)**	**4 (7.4)**	**4 (7.4)**	**3 (5.5)**	**2 (3.7)**	**1 (1.9)**	**1 (1.9)**	**1 (1.9)**	**1 (1.9)**	**54 (100)**

Abbreviations : CHS: chondrosarcoma; CS: carcinosarcoma; DFSP: dermatofibrosarcoma protuberans; ETH B: ethmoid bone; EWS: Ewing Sarcoma; KS: Kaposi sarcoma ; LMS: leiomyosarcoma; Mand : mandible ; Max: maxilla ; MFH : malignant fibrous histiocytoma; MPNST: Malignant peripheral nerve sheath tumor; MSFT: Malignant solitary fibrous tumor; NPH: nasopharynx; OC: oral cavity; OSS: osteosarcoma; RMS: rhabdomyosarcoma; SNC: sinonasal cavity; UDS: undifferenciated sarcoma

Table 3: Comparison of demographic and topographic characteristics in the present study with other reported series

	Stavrakas (UK) [5]	*Alishahi (Iran) [3]*	*Hammami (Tunisia) [6]*	*Bobby (USA) [7]*	*Piñeiro Aguín (Spain) [12]*	*Pacheco (Brazil) [13]*	*Rapidis (Greece) [9]*	*Barosa (Portugal) [4]*	*Borki (Morocco) [2]*	*Present series*
SR M/F	2.54*	1.14*	0.66	1.38*	1.77*	1.76	1:1.5	2.22*	2.23*	1.5 :1
Mean age (range) yrs	- (13-88)	41 (1 – 85)	38,4 (6-73)	49±22 -	- (1-78)	39.7 (20-59)	42.5 (16-76)	45.9 ± 25 (5-90)	35.5 (13-70)	37.7 (1-88)
Location %										
- mand	10.2	NS	0	7.0	0	-	NS	4/29	28.6	7.4
- Maxilla	0	NS	0	8.1	0	-	NS	-	9.5	5.5
- ETH B	0	NS	0	-	0	0	NS	0	0	1.9
- Face	-	NS	0	10.8	0	22.2	NS	0	-	22.2
- scalp	-	NS	0	12.4	0	19.4	NS	0	0	14.8
- SNC	23	NS	20	22	32	-	NS	41.3*	4.8	11.1
- Orbit	0	NS	0	12.9	0	13.8*	NS	0	2.4	12.9
-Larynx	12.8	NS	13.3*	-	14	-	NS	13.8*	2.4	7.4
-NPH	-	NS	6.6	-	0	0	NS	3.4*	9.5	5.5
-Parotid	-	NS	6.6	-	0	0	NS	0	11.9	3.7
- OC	15.3	NS	20	2.2	8	13.8	NS	20.7*	7.2*	1.9
-Ear	0	NS	0	-	4	0	NS	0	0	1.9
-Neck	-	NS	13.3*	14	32	22.2	NS	0	2.4	1.9
- Thyroid	-	NS	13.3*	-	0	-	NS	0	0	1.9
-OPH	10.2	NS	0	-	8	0	NS	0	4.8	0
-PPHS	0	NS	6.6	-	0	1/36	NS	0	2.4	0
-Others	NS	NS	0	18.8	0	0	NS	6.9*	14.3*	0
Total	**39**	**105**	**15**	**186**	**25**	**36**	**25**	**29**	**42**	**54**

* Value that we calculated from the data presented in the article

Abbreviations: CHS: ETH B: ethmoid bone; F: female; M: male; Mand: mandible; NPH: nasopharynx; NS: not specified; OC: oral cavity; OPH: oropharynx; PPHS: parapharyngeal space; SNC: sinonasal cavity; SR: sex-ratio; yrs: years

Table 4: Comparison of histopatholpogical characteristics in the present study with other reported series

	Vassiliou (UK) [1] n (%)	*Stavrakas (UK) [5] n (%)*	*Alishahi (Iran) [3] n (%)*	*Hammami (Tunisia) [6] n (%)*	*Bobby (USA) [7] n (%)*	*Piñeiro Aguín (Spain) [12] n (%)*	*Pacheco (Brazil) [13] n (%)*	*Ketabchi (UK) [14] n (%)*	*Rapidis (Greece) [9] n (%)*	*Barosa (Portugal) [4] n (%)*	*Borki (Morocco) [2] n (%)*	*Present series n (%)*
OSS	41	4 (10.2)	32(30.47)	1 (6,6)	25(13.4)	0	2 (5,6)	17	5	10 (34)	8 (19)	4 (7.4)
EWS	5	1 (2.5)	11(10.47)	0	4 (2.2)	0	1 (2,8)	*2*	1	0	1 (2.4)	1 (1.9)
CHS	8	6 (15.3)	14(13.33)	2 (13,3)	16 (8.6)	3 (12)	0	1	3	2 (7)	4 (9.5)	4 (7.4)
GCS	0	0	0	0	1 (0.5)	0	0	0	0	0	0	0
RMS	12	1 (2.5)	8 (7.61)	5 (33,3)	19(10.2)	4 (16)	9 (25)	1	1	5 (17)	7 (16.7)	15(27.7)
SCS	6	3 (7.6)	3 (2.85)	0	0	0	1 (2,8)	0	0	0	0	0
MFS	5	0	0	0	3 (1.6)	0	2 (5,6)	1	0	0	0	0
SS	3	2 (5.1)	6 (5.71)	0	5 (2.7)	6 (24)	0	1	0	1 (3)	14(33.3)	0
MPNST	4	0	8 (7.61)	1 (6,6)	13 (7)	1 (4)	2 (5,6)	0	0	3 (10)	0	1 (1.9)
MyFS	3	1 (2.5)	0	0	1 (0.5)	0	0	0	0	0	0	0
LMS	5	3 (7.6)	0	2 (13,3)	12(6.45)	2 (8)	0	1	4	3 (10)	3 (7.1)	10(18.5)
LPS	2	1 (2.5)	3 (2.85)	0	6 (3.2)	1 (4)	3 (8,3)	0	1	0	1 (2.4)	0
DFS	2	3 (7.6)	0	0	1 (0.5)	0	5 (13,8)	0	1	0		3(5.5)
AS	1	2 (5.1)	4 (3.80)	0	18(9.67)	0	0	0	1	3 (10)	1 (2.4)	0
SFT-HPC	0	0	0	0	41 (22)	0	0	0	0	0	0	2(3.7)
CS	0	2 (5.1)	0	0	0	0	2 (5,6)	0	0	0	0	7(13)
Alveolar soft part	1	0	0	0	0	0	0	1	0	0	0	0
MFH/ UDS/PS	9	2 (5)	4 (3.80)	2 (13,3)	18 (9.7)	7 (28)	4 11.1)	0	5	3 (10)	2 (4.8)	1 (1.9)
KS/NSS	0	8 (20.5)	2 (1.90)	0	2 (1.1)	1 (4)	NI	0	1	3 (10)	0	5(9.2)
FS	0	0	6 (5.71)	2 (13,3)	0	0	5 (13,8)	0	2	0	1 (2.4)	0
AFX	0	0	0	0	1 (0.5)	0	0	0	0	0	0	0
Metastatic sarcoma	0	0	4 (3.80)	0	0	0	NI	0	0	0	0	0
Total	**107**	**39**	**105**	**15**	**186**	**25**	**36**	**25**	**25**	**29**	**42**	**54**

AFX : atypical fibroxanthoma ; AS : angiosarcoma ; CS: carcinosarcoma; DFS : dermatofibrosarcoma ; FS: fibrosarcoma; GCS : giant cell sarcoma ; KS: Kaposi sarcoma ; LPS : liposarcoma ; MFH : malignant fibrous histiocytoma ; MFS : Myxofibrosarcoma ; MPNST : Malignant peripheral nerve sheath tumor ; MyFS : myofibrosarcoma ; NSS : non specified sarcoma ; PS : pleomorphic sarcoma ; SCS : Spindle cell sarcoma ; SFT-HPC : Solitary fibrous tumor/hemangiopericytoma ; SS : Synovial sarcoma ; UDS: undifferenciated sarcoma

DISCUSSION

Sarcomas of the head and neck are very rare, accounting for approximately 1% of all head and neck neoplasms [7]. In Morocco, few papers have been published [2] and this is the first survey conducted in the region of Fez.

Analysis of gender repartition in the present study showed a higher frequency in males. This finding is consistent with that of most studies [2-5, 7, 9, 11-13]. Head and neck sarcomas were rarely more common in females [6].

Concerning age, this study revealed a younger age. In fact, mean age was 37.7 years whereas most reviewed series reported a higher mean age [3, 4, 6, 7, 9, 13]. Another Moroccan study found, however, a lower average age (31.6 years) [2]. Analysis of distribution by decade showed, in the study published by Rapidis [9], a peak at the third decade, the same found herein.

Repartition into two groups, adults and children; found that the formers were about three times more affected than the latter. Children proportion were lower in the analysis of the SEER Database that found, among a total of 12,725 cases of head and neck sarcoma, 1244 pediatric patients [11]. Comparison of epidemiological characteristics in the present study with other reported series is presented in table 3.

Comparison of topographic distribution of tumors was challenging because of the heterogeneity of adopted topographic repartition between studies. In this survey, a higher proportion of soft tissue sarcomas were recorded (83.3%) in comparison with hard tissue sarcomas (16.7%). In the region of Rabat, the same result was reported [2]. Alishahi [3] and Ketabchi [14] reported an adverse result with hard tissue sarcomas being more frequent than the soft tissue ones. In the results published by Vassiliou [1], soft tissue sarcomas were as frequent as bone ones (53 and 54 cases respectively). In the literature, most series indicated sinonasal cavity as the commonest location [4-7, 12], followed by oral cavity in some series [4, 5] and by neck in others [7]. Sinonasal location was as frequent as oral cavity in the study reported by Hammami [6] and as neck in the study reported by Piñeiro Aguín [12]. Result was different in the study conducted by Pacheco [13] in which face and neck were the most frequent locations or in the study by Borki [2] in which mandible and parotid were the commonest sites. In this survey, the most common locations were face and scalp. Sinonasal cavity was the fourth affected site. Table 3 provides results of topographic repartition in previous published series and in ours.

When considering only hard tissue sarcomas, mandible was the most common, followed by maxilla. This result is in accordance with that found by Vassiliou who noticed a proportion of mandibular location of 54%, followed by the maxilla (34%), whereas 11% of bone sarcomas occurred in extragnathic locations [1].

On histopathological analysis, rhabdomyosarcoma and leiomyosarcoma were the top two histological types in this study (27.7% and 18.5% respectively), followed by carcinosarcoma, Kaposi sarcoma, chondrosarcoma, osteosarcoma. Other types were very rare. In the literature, results differed according to series. Osteosarcoma was frequently reported as the most preponderant type [1, 3, 4, 9, 14]. In this survey, osteosarcoma was the fifth most frequent tumor and represented only 7.4% of all head and neck sarcomas. Rare studies shared the same result as the present series and noticed that rhabdomyosarcoma was the commonest type [6, 13]. Other studies reported variable results with Kaposi sarcoma [5], solitary fibrous tumor [7] or synovial sarcoma [2, 12] reported as the most frequent. Concerning leiomyosarcoma, it was rarely classified among the two most frequent sarcomas [9]. Histological repartition in the present and previous series is detailed in table 4.

The tumor spectra varied from adults to children. In fact, osteosarcoma is the most common sarcoma in the head and neck region among adults, and rhabdomyosarcoma is the most common in children [4, 15]. This pediatric finding was confirmed by the present study whereas adult result was different with leiomyosarcoma being the most frequent.

When the sex distribution in different histological types was compared, it was found that leiomyosarcoma and carcinosarcoma were more frequent in males whereas chondrosarcoma and dermatofibrosarcoma protuberans were more frequent in females. Rhabdomyosarcoma, Kaposi sarcoma and osteosarcoma were equally seen in both sexes. In the literature, results were different. Concerning rhabdomyosarcoma, it occurred commonly in girls [3, 16-18], unlike the results from study by Chidzonga [19] in which the disease was prominent in boys.

Concerning bone tumors, Alishahi [3] showed a male preponderance for osteosarcoma and an equal gender distribution of chondrosarcoma. But in the study conducted by Guevara Canales [18], osteosarcoma and chondrosarcoma were found mainly in females.

CONCLUSION

This study confirms rarity and heterogeneity of head and neck sarcomas. In Fez, these tumors share some characteristics with previous published series and present some differences. They occur mostly in males,

with a younger age at diagnosis and a higher proportion of children. Soft tissue location is the most frequent, particularly facial and rhabdomyosarcoma and leiomyosarcoma are the commonest types. This study was limited by the fact that the sample was heterogeneous conducting to a small number of each type. A larger series is necessary to better characterize all histological categories.

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