ORIGINAL ARTICLES

Incidence, and Gender, Age and Ethnic Distribution of Sarcomas in the Republic of Suriname from 1980 to 2008

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ABSTRACT

Objective: *We report on the incidence and the gender, age and ethnic distribution of sarcomas diagnosed between 1980 and 2008 in the multi-ethnic Republic of Suriname.*

Methods: *Total and average yearly number of cases, crude rates, as well as relevant population data were derived from the records of the Pathologic Anatomy Laboratory and the General Bureau of Statistics, respectively, and stratified according to gender, age groups 0–19, 20–49 and 50+ years, and the largest ethnic groups (Hindustani, Creole, Javanese and Maroons).*

Results: *Between 1980 and 2008, 258 sarcomas were diagnosed in Suriname,* ie *at a frequency of nine per year and an annual rate of two per 100 000. Overall, there was 0.9 male per female, two to four cases per year in each age group, and one to three patients in each ethnic group. Soft-tissue sarcomas comprised approximately 80% of overall cases, with a male/female ratio that was approximately 0.5; almost 90% of patients were older than 20 years; more than one-third was Creole. Leiomyosarcoma, fibrosarcoma and liposarcoma were most frequently encountered (90 cases), particularly above 20 years of age, while leiomyosarcomas seemed, additionally, more common in women and Creoles or Maroons. The most numerous bone tumours were primitive neuroectodermal tumour/Ewing tumour and osteosarcoma (37 cases). They were more common in males, the youngest age group, and Hindustanis and Creoles.*

Conclusions: *The incidence of sarcomas in Suriname, and their gender, age and ethnic distribution in general, seemed comparable with international data. The main exception might be leiomyosarcoma which might have a predilection for Afro-Surinamese.*

Keywords: Age distribution, ethnic distribution, gender distribution, incidence, sarcomas, Suriname

Incidencia y Distribución de los Sarcomas por Género, Edad y Etnia en la República de Surinam desde 1980 a 2008

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RESUMEN

Objetivo: *Reportamos la incidencia y distribución de los sarcomas por género, edad y etnia, diagnosticados en la población multiétnica de la República de Surinam entre 1980 y 2008.*

Métodos: *El total y el promedio anual del número de casos, tasas brutas, así como los datos relevantes de la población fueron obtenidos de los registros del Laboratorio de Anatomía Patológica y la Dirección General de Estadísticas respectivamente, y estratificados según el género, los grupos etarios de 0–19, 20-49 y más de 50 años de edad, y los grupos étnicos mayores (indostánicos, criollos, javaneses y cimarrones).*

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Resultados: *Entre 1980 y 2008, se diagnosticaron 258 sarcomas en Surinam, lo que equivale a una frecuencia de nueve por año y una tasa anual de dos por cada 100 000. En general, hubo 0.9 varones por hembras, de dos a cuatro casos por año en cada grupo etario, y de uno a tres pacientes en cada grupo étnico. Los sarcomas de tejidos blandos constituyeron aproximadamente el 80% de la generalidad de los casos, con una proporción varón/hembra de 0.5 aproximadamente; casi el 90% de los pacientes tenía más de 20 años; más de un tercio eran criollos. Los leiomiosarcomas, fibrosarcomas y liposarcomas fueron los más frecuentes (90 casos), particularmente pasados los 20 años de edad, presentándose además los leiomiosarcomas más comúnmente en las mujeres y en los criollos o los cimarrones.*

Palabras claves: Distribución por edad, distribución étnica, incidencia, sarcomas, distribución por género, Surinam

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INTRODUCTION

Sarcomas comprise a heterogeneous group of malignant neoplasms of mesenchymal origin. Because mesenchymal tissues are found anywhere in the body, these malignancies can occur in any organ or organ system (1). However, they are commonly seen in extremities, trunk and pelvis and the head and neck (1). They are usually treated with surgery, chemotherapy (particularly for Ewing's and rhabdomyosarcoma (2)) and biological therapies aimed at mutated tumour-specific oncogenes (3). Unfortunately, these therapeutic modalities produce cure rates of at the most 30% (4), principally because sarcomas are commonly diagnosed at an advanced stage (1, 4).

The development of certain soft-tissue sarcomas has been associated with a number of inherited disorders such as Li-Fraumeni syndrome and neurofibromatosis (5, 6). A few other cases have been associated with exposure to radiation (for instance, in the form of radiation therapy given for breast cancer or lymphoma (7)), as well as high doses of vinyl chloride (8), dioxin (9), and herbicides that contain phenoxyacetic acid (10). However, with the notable exception of Kaposi's sarcoma – caused by the human herpes virus 8 (11) – the exact causes of most sarcomas are still unknown.

International incidence rates are estimated to range between 1.8 and 5.0 cases per 100 000 per year for soft-tissue sarcomas (12, 13), and between 0.03 and 0.3 cases per 100 000 per year for bone sarcomas (14, 15). There may be a slight male predominance, with a male-to-female ratio of 1.1 : 1.0, but this may vary among histologies (14, 16, 17). Furthermore, they are much more common in children than in adults, accounting each year for approximately 15% of new cancer cases in the former group, and roughly 1% in the latter (12, 14, 16, 17).

The Republic of Suriname in South America harbours an ethnically highly diverse population of about 531 000 that includes Hindustanis and Javanese, originating from India and Java (Indonesia), respectively; Creoles, from mixed white and black ancestry; Maroons, the immediate descendants from runaway African slaves; people from Chinese, European, Middle Eastern, and Brazilian descent, as well as Amerindians, the original inhabitants (18).

We recently reported on various aspects of the epidemiology of cancer in Suriname (19, 20). So far, however, no comprehensive studies have been carried out on the occurrence of sarcomas in the country. Therefore, and considering the peculiar epidemiological aspects of this group of malignancies, we decided to assess them for their incidence in the uniquely varied population of Suriname. The period of time covered by this study was 1980 to 2008. The data obtained have been stratified according to gender, age, and ethnic background, and have been discussed against the background of global data on the epidemiology of sarcomas.

SUBJECTS AND METHODS

In this survey, the histopathologically confirmed sarcomas diagnosed in Suriname between January 1, 1980 and December 31, 2008 have been inventoried and stratified on the basis of histopathology, gender, age at the time of diagnosis, and ethnic background. Benign neoplasms of mesenchymal origin, *in situ* malignancies, and skin cancers have been excluded from this study.

Numbers of diagnoses of sarcoma in the period covered by this study were obtained from patients' records at the Pathologic Anatomical Laboratory of the Academic Hospital Paramaribo. This institution is the reference centre for the histopathological diagnosis of cancer and the registration of histopathologically diagnosed cancer cases in Suriname. All cases have been classified according to the recommendations of the World Health Organization (21). The records of the Pathologic Anatomical Laboratory also provided details about the histology of the malignancies, as well as information about gender, age at the time of diagnosis, and ethnic background of the patients.

Population data, including estimates of the total mid-year resident population size of Suriname, as well as the male and female resident population size from 1980 to 2008, were provided by the Department of Population Statistics of the General Bureau for Statistics, Ministry of Planning and Developmental Cooperation (18).

For each year in the period 1980 to 2008, the total number of sarcomas, the total number of soft-tissue and bone tumours, the total number of each histology, the total number of male and female cases, the total number of cases in age groups 0 to 19 years, 20 to 49 years, and 50 years and older, as well as the total number of cases in the four largest ethnic groups (Hindustani, 37%; Creoles, 31%; Javanese, 14.6% and Maroons, 14%) has been determined.

For each (sub) stratum, average yearly number of cases and average yearly crude and gender-specific rates have been calculated. The latter was done by dividing the number of cases in each (sub-) stratum by either the estimated total mid-year resident population or the estimated mid-year male or female population, and were expressed per 100 000 population, or per 100 000 men or women, respectively, per year.

Yearly frequencies and crude incidence rates are presented. The latter have been expressed as means ± standard deviations (SDs) and have been compared by taking p-values < 0.05 to indicate statistically significant differences according to analysis of variance (ANOVA) and Fisher's exact test.

RESULTS

From January 1980 to December 2008, 258 cases of sarcoma were diagnosed in Suriname (Table 1). This corresponded to approximately nine new cases per year, and a crude rate of about two per 100 000 individuals per year. The male-to-female ratio was 0.9 (about 47% male and 53% female patients). The tumours presented in both genders at a rate of four to five per year, or about one per 100 000 per year (Table 1).

Table 1: Characteristics of patients with sarcomas in Suriname, 1980–2008. Crude rates are 100 000 population per year

	Number of cases	% of total	Average yearly number of cases ± SD	Crude rate ± SD
Males	121	46.9	4.2 ± 1.9	9.9 ± 4.5
Females	136	52.7	4.7 ± 2.4	11.0 ± 5.3
Unknown	2	0.8	–	–
0–19 years	56	21.7	2.0 ± 1.5	4.7 ± 3.7
20–49 years	101	39.1	3.5 ± 2.0[1]	8.2 ± 4.7[1]
50+ years	92	35.7	3.2 ± 2.0[2]	7.3 ± 4.1[2]
Unknown	9	2.6	–	–
Hindustanis	59	22.9	2.0 ± 1.5[3]	4.7 ± 3.4[3]
Creoles	95	36.8	3.3 ± 1.8[4]	7.8 ± 4.1[4]
Javanese	47	18.2	1.6 ± 1.4	3.9 ± 3.4
Maroons	24	9.3	0.8 ± 0.9	1.9 ± 2.1
Others/Unknown	33	12.8	1.1 ± 1.0	2.7 ± 2.4
Total	**258**	**100**	**8.9 ± 3.3**	**21.0 ± 7.0**

[1]Significantly different from '0–19 years' ($p < 0.01$, ANOVA); [2]significantly different from '0–19 years' ($p < 0.05$, ANOVA); [3]significantly different from 'Maroons' ($p < 0.01$, ANOVA); [4]significantly different from 'Hindustanis', 'Javanese', Maroons' and 'Others/Unknown' ($p < 0.01$, ANOVA)

The median age of the patients was 43 years, ranging from 0 to 89 years. About 20% of the patients were between 0 and 19 years of age; about 40% was between 20 and 49 years, and a similar proportion was 50 years and older (Table 1). There were on average two, four and three cases per year in each of these age groups. Approximately one-third of the patients were Creole. This was 1.5 to 4 times higher than the number of patients with a Hindustani, Javanese, or Maroon background. These values corresponded with roughly three Creole, two Hindustani, one to two Javanese, and one Maroon patient(s) presenting each year with a sarcoma (Table 1).

As shown in Table 2, about 80% of the sarcomas diagnosed in the period covered by this study comprised soft-tissue sarcomas while about 20% were bone tumours. The malignancies were encountered in all parts of the body. However, almost half (45%) was located in the trunk, 25% in the limbs, 5% had affected the head and neck region and 2% the spine (Table 2).

The most frequently diagnosed soft-tissue sarcomas between the years 1980 and 2008 were leiomyosarcoma (37 cases), fibrosarcoma (29 cases), liposarcoma not otherwise specified [NOS] (24 cases), pleomorphic undifferentiated sarcoma (20 cases), Kaposi's sarcoma (13 cases) and undifferentiated sarcoma/NOS (35 cases). Together, they comprised roughly two-thirds of the 258 sarcomas registered in the period covered by this study. The overall male-to-female ratio was about 2:3 (Table 3). Furthermore, almost 90% of patients were older than 20 years, and there were similar numbers of patients aged 20 to 49 years to those 50 years and older (Table 3). In addition, more than one-third of the patients were Creole, 22% Hindustani, 20% Javanese and 9% Maroon.

Leiomyosarcoma comprised approximately one-quarter of the most common soft-tissue sarcomas (Table 3). The majority of these patients were female (31 out of 37), 50 years or older (24 out of 37), and either Creole or Maroon (12 and nine, respectively, out of 37). Fibrosarcoma, liposarcoma, pleomorphic undifferentiated sarcoma, and Kaposi's sarcoma represented roughly one-tenth to one-fifth of the most common soft-tissue sarcoma (Table 3). With the exception of Kaposi's sarcoma (that was twice more common in men than in women), these tumours seemed to occur as often in either genders (Table 3). However, the vast majority (75 to 100%) was encountered in individuals 20 years and older (Table 3), while about half of fibrosarcomas and almost all Kaposi's sarcomas were in Creoles (Table 3).

In the period covered by this study, there were, at the most, nine cases of myxofibrosarcoma, rhabdomyosarcoma, synovial sarcoma, angiosarcoma, dermatofibrosarcoma protuberans, desmoplastic small round cell tumour, clear cell sarcoma and alveolar soft part sarcoma (Table 4). Together, these soft-tissue sarcomas comprised approximately one-fifth of the total number of sarcomas seen in this period, seemed to lack a clear male-to-female preference, occurred three

Table 2: Primary sites of sarcomas in Suriname between 1980 and 2008. Percentages of the number of overall sarcomas are given in brackets

	Limbs	Head and neck	Trunk	Spine	Unknown	Total
Soft-tissue sarcomas	52 (20)	13 (5)	113 (44)	–	31 (12)	210 (81)
Bone sarcomas	13 (5)	1 (0)	2 (1)	4 (2)	28 (11)	48 (19)
Total	65 (25)	14 (5)	115 (45)	4 (2)	59 (23)	258 (100)

Table 3: Characteristics of most common soft-tissue sarcomas in Suriname between the years 1980 and 2008

	Total number	Male-to-female ratio	Age group (years)			Ethnic background			
			0–19	20–49	50+	Hindustanis	Creoles	Javanese	Maroons
Leiomyosarcoma, NOS (M 8890/3)	37	6/31	2	9	24	7	12	5	9
Fibrosarcoma (M 8810/3)	29	13/16	4	16	9	3	13	4	3
Liposarcoma, NOS (M 8850/3)	24	13/11	0	13	11	10	9	4	0
Pleomorphic undifferentiated sarcoma (M 8830/3)	20	12/8	2	8	7	4	5	7	0
Kaposi's sarcoma (M 9140/3)	13	9/4	1	3	9	0	12	0	1
Undifferentiated sarcoma/NOS (M 8800/3)	35	8/27	4	23	9	11	8	12	2
Total	**158**	**61/97**	**13**	**72**	**69**	**33**	**59**	**32**	**15**

NOS: not otherwise specified

Table 4: Characteristics of less common sarcomas in Suriname between the years 1980 and 2008

	Total number	Male-to-female ratio	Age group (years)			Ethnic background			
			0–19	20–49	50+	Hindustanis	Creoles	Javanese	Maroons
Myxofibrosarcoma (M 8811/3)	9	7/2	0	0	9	2	6	1	0
Rhabdomyosarcoma (M 8910/3	9	7/2	8	1	0	2	2	4	0
Synovial sarcoma (M 9040/39)	9	7/2	0	7	2	2	3	1	1
Angiosarcoma (M 9120/3)	6	2/4	0	2	4	1	3	0	1
Dermatofibrosarcoma protuberans (M 8832/3)	6	2/4	0	6	0	1	3	0	2
Desmoplastic small round cell tumour (M 8806/3)	5	3/2	3	0	2	0	4	0	0
Clear cell sarcoma (M 9044/3)	5	2/3	1	2	2	1	0	1	1
Alveolar soft part sarcoma (M 9581/3)	3	1/2	1	2	0	2	1	0	0
Total	**52**	**31/21**	**13**	**20**	**19**	**11**	**22**	**7**	**5**

times more often in individuals 20 years and older than in those younger than 20 years, and were at least twice more common in Creoles than in the other ethnic groups (Table 4).

However, myxofibrosarcoma, rhabdomyosarcoma and synovial sarcoma might have a predilection for males rather than females (Table 4). Also, myxofibrosarcoma, synovial sarcoma, angiosarcoma, dermatofibrosarcoma protuberans, and clear cell sarcoma seemed to occur more often at age 20 years and older, while rhabdomyosarcoma was clearly more prevalent in the age group 0 to 19 years (Table 4). Furthermore, despite the relatively small numbers of cases, particularly myxofibrosarcoma and desmoplastic small round cell tumour might be more common in Creoles than in the other ethnic groups (Table 4).

There were 19, 18 and 11 cases of primitive neuroectodermal tumour (PNET)/Ewing tumour, osteosarcoma, and chondrosarcoma, respectively, in the period covered by this study (Table 5). For all three histologies, there were more male than female patients. Approximately one-quarter of overall bone sarcomas was seen in individuals aged between 20 and 49 years, but about two-thirds in those between 0 and 19 years of age. Accordingly, 60 to 80% of cases of osteosarcoma and PNET/Ewing tumour were in the youngest age group. Overall, bone sarcomas were most frequently seen in Hindustanis and Creoles (approximately 30% in each of the groups) and were least common in Maroons (only 4%). Markedly, there were no cases of PNET/Ewing tumour in the latter ethnic group (Table 5).

Table 5: Characteristics of bone sarcomas in Suriname between the years 1980 and 2008

	Total number	Male-to-female ratio	Age group (years)			Ethnic background			
			0–19	20–49	50+	Hindustanis	Creoles	Javanese	Maroons
PNET/Ewing tumour (M 9364/3)	19	16/3	15	4	0	8	6	4	0
Osteosarcoma (M 9180/3)	18	11/7	11	3	4	5	7	1	1
Chondrosarcoma (M 9240/3)	11	7/4	5	4	2	1	3	2	1
Total	**48**	**34/14**	**31**	**11**	**6**	**14**	**16**	**7**	**2**

PNET: primitive neuroectodermal tumour

DISCUSSION

Due to their rarity and heterogeneity, the epidemiology – and hence the aetiology – of sarcomas is still incompletely understood. In this descriptive study, we attempted to gain more insight into this subject by assessing this group of malignancies by overall occurrence and their gender, age, and ethnic distribution in the multi-ethnic Republic of Suriname. In the period between 1980 and 2008, 258 such malignancies were diagnosed in the country. This corresponded to approximately nine cases per year. Considering the previously reported average incidence in Suriname of roughly 350 new malignant neoplasms per year (20), sarcomas can thus be estimated to comprise around 2% of these conditions. This is fairly well in the range of estimated incidence rates for these malignancies of 1 to 3% in most parts of the world (12, 14, 16, 17).

The four-fold greater number of soft-tissue sarcomas when compared to bone sarcomas noted in the present study (81 *versus* 19%) is in accordance with the occurrence of 11 280 soft-tissue sarcomas and 2890 bone sarcomas in the United States of America (USA) in the year 2012 (4). The presence of the majority of the primary sites in the trunk and limbs (45 and 25%, respectively) is in accordance with the observation that soft-tissue sarcomas originate rather frequently within the abdomen or in the limb or limb girdle, both retroperitoneally or viscerally and intraperitoneally (22).

The male-to-female ratio of 0.9, and the greater number of older patients when compared to younger ones noted in the present study, are also in agreement with the practically equal number of male and female sarcoma patients throughout the world (4, 14, 16, 17), and the generally higher prevalence of neoplastic disease in individuals older than 50 years when compared to younger ones (4, 14, 16, 17). The apparent predominance of Creole patients with a sarcoma over those from Hindustani, Javanese, or Maroon background cannot be satisfactorily explained, but is in line with the occurrence of more malignancies in the former group when compared to the latter three (19, 20).

Despite the relatively small number of cases, the gender, age, and ethnic distribution of the most common soft-tissue sarcomas encountered in this study (leiomyosarcoma, fibrosarcoma, liposarcoma, pleomorphic undifferentiated sarcoma, Kaposi's sarcoma and undifferentiated sarcoma/NOS) was essentially as expected. The far greater number of leiomyosarcomas in females when compared to males might be attributed to the presumed stimulatory effect of oestrogen on smooth muscle proliferation (23). On the other hand, in accordance with previous observations (24–26), there was no

clear gender predilection for fibrosarcoma, liposarcoma and pleomorphic undifferentiated sarcoma.

The presentation of most patients with leiomyosarcoma, pleomorphic undifferentiated sarcoma, or liposarcoma at age 50 years and older, and with fibrosarcoma in the age group between 20 and 49 years, is also in agreement with previously published data (24–27). However, leiomyosarcomas, fibrosarcomas and Kaposi's sarcomas seemed to occur more frequently in Creoles and Maroons than in Hindustanis and Javanese. These malignancies have so far not been associated with a racial predilection, making it difficult to explain our observations satisfactorily. However, if true, they suggest that Afro-Surinamese may be either more susceptible to these malignancies or may be more exposed to associated risk factors when compared to Asian-Surinamese. These assumptions need to be verified in future studies.

The numbers of myxofibrosarcomas, rhabdomyosarcomas, synovial sarcomas, angiosarcomas, dermatofibrosarcomas protuberantes, desmoplastic small round cell tumours, clear cell sarcomas and alveolar soft part sarcomas were even smaller than those of the sarcomas previously mentioned. This makes suggestions about their gender, age and ethnic distribution even more difficult. Nevertheless, our observations seem consistent with the absence of an obvious gender predilection of these malignancies (14, 16, 17), as well as with the preferential occurrence of myxofibrosarcoma in older individuals (28) and rhabdomyosarcoma in children (29).

The 48 cases of PNET/Ewing's tumour, osteosarcoma, and chondrosarcoma seen in the 29 years covered by this study suggest that bone cancers occurred in Suriname at an average frequency of one to two cases per year, or roughly 0.3 per 100 000 population per year. This is within the range of 0.03 to 0.3 per 100 000 per year mentioned for other parts of the world (15). The consistent male-over-female prevalence of these tumour types seen in Suriname is also in accordance with international trends (15).

The same holds true for the observation that PNET/Ewing's sarcomas and osteosarcomas were mostly found in individuals younger than 20 years. This is completely in agreement with the occurrence of these malignancies around the age of 14 years, coinciding with the pubertal growth spurt (30, 31). The distribution of either PNET/Ewing's tumour and osteosarcoma, or that of chondrosarcoma displayed apparent ethnic preferences. However, the absence of Maroons with Ewing's sarcoma is in line with the rarity of this tumour type in individuals from African descent when compared to Asians and Caucasians (32).

Summarizing, in line with international data, sarcomas were also very rare in Suriname, comprising 2 to 3% of all malignant neoplasms diagnosed in the country, with approximately four times more soft-tissue sarcomas than bone tumours. In general, there were no obvious peculiarities with regard to their gender, age and ethnic distribution. However, individuals from African descent may be more susceptible to soft-tissue sarcomas, particularly leiomyosarcoma, when compared to those with an Asian background. These suggestions need to be further assessed in larger studies examining patient samples for relevant molecular markers and employing sufficient statistical power.

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