Epidemiologic Data on Meningiomas in Jamaica: The First from the Caribbean

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Citation

Abstract
There is to date no data from the Caribbean regarding the occurrence of meningiomas. This ten year retrospective study examines the epidemiological features of meningiomas at the University Hospital of the West Indies, Jamaica. All meningiomas which were resected at this institution during the study period as well as those discovered at autopsy were included. There were a total of 67 meningiomas. 90% were surgically resected and of these 12% were spinal in location and 88% were intracranial with a male: female ratio of 1:7 and 1:2.8 respectively. Of the intracranial meningiomas, 89% were within the supratentorial compartment. Six percent of the total was malignant (WHO Grade III). Of the 7 documented recurrences, 2 were malignant, 2 were incompletely resected and 1 showed brain invasion histologically. This study is the first to reveal the epidemiological pattern of meningiomas in Kingston, Jamaica and also highlights some important aspects of those which recur.

INTRODUCTION
Meningiomas are neoplasms arising from meningothelial cells which occur in greatest abundance in the arachnoid villi but are also encountered as small clusters throughout the craniospinal space. They have probably affected humans since prehistoric times. The term meningioma was popularized by Harvey Cushing in the early 1900's (1).

The majority of these tumors are benign and slow growing and it is thought that most meningiomas have been present for a decade or more at the time of diagnosis. They may present with a variety of symptoms and signs depending on their size and location.

Numerous articles have been written over the years from all over the world on various aspects of these lesions; however, information from the Caribbean has been sparse. The present study, the first of its kind in regards to meningiomas, was therefore designed to examine the epidemiological profile of surgically resected and autopsy detected meningiomas at the University Hospital of the West Indies (UHWI), Kingston, Jamaica over a ten year period, 1994 to 2003. A comparative review of these figures with international data is presented along with a brief general review of the epidemiology, biology and classification of this tumor.

MATERIALS AND METHODS
The University Hospital of the West Indies (UHWI) is a tertiary care teaching hospital located in Kingston, the capital of the island of Jamaica in the West Indies. All meningiomas entered in the files of the Pathology Department at the UHWI between January 1994 and December 2003 was reviewed. The material was derived from surgically resected specimens done at the UHWI as well as from autopsies performed at this institution within that period.

Post mortem cases in which the diagnosis had already been established by histopathology were excluded, as were all cases not derived from the Neurosurgical Unit of the UHWI. As the specimens received from other hospitals have not been included, the results therefore do not necessarily reflect the occurrence of meningiomas in the island as a whole.

Classification of all the meningiomas was done according to the standard World Health Organization (WHO) method, 1993.

RESULTS
During the study period, a total of 67 meningiomas were identified. Of these, 60 (90%) were sent to the Department as surgical resection specimens and 7(10%) were entered into the records as autopsy specimens (Table 1).
Epidemiologic Data on Meningiomas in Jamaica: The First from the Caribbean

Figure 1
Table 1: Source and Topographical Distribution of Meningiomas at U.H.W.I (1994-2003)

<table>
<thead>
<tr>
<th>Type</th>
<th>Intracranial</th>
<th>Spinal</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td>Surgical</td>
<td>59</td>
<td>7</td>
<td>66</td>
</tr>
<tr>
<td>Postmortem</td>
<td>7</td>
<td>0</td>
<td>7</td>
</tr>
<tr>
<td>Total</td>
<td>66</td>
<td>7</td>
<td>73</td>
</tr>
</tbody>
</table>

Of the total number of meningiomas encountered during this time period, the majority were seen in the 31 to 60 year age group which accounted for 73% of the total. The 41 to 50 year age group was the single group in which most meningiomas occurred (Table 2).

Figure 2
Table 2: Age Distribution of Meningiomas at the UHWI (1994-2003)

<table>
<thead>
<tr>
<th>Age (Year)</th>
<th>0-19</th>
<th>20-29</th>
<th>30-39</th>
<th>40-49</th>
<th>50-59</th>
<th>60-69</th>
<th>70-79</th>
<th>TOTAL</th>
</tr>
</thead>
<tbody>
<tr>
<td>IC* (Resected)</td>
<td>0</td>
<td>2</td>
<td>3</td>
<td>16</td>
<td>14</td>
<td>10</td>
<td>3</td>
<td>35</td>
</tr>
<tr>
<td>Sp** (Resected)</td>
<td>0</td>
<td>1</td>
<td>0</td>
<td>1</td>
<td>1</td>
<td>2</td>
<td>2</td>
<td>7</td>
</tr>
<tr>
<td>Autopsy</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>4</td>
<td>1</td>
<td>1</td>
<td>1</td>
<td>7</td>
</tr>
<tr>
<td>Total</td>
<td>0</td>
<td>3</td>
<td>2</td>
<td>21</td>
<td>12</td>
<td>8</td>
<td>5</td>
<td>67</td>
</tr>
</tbody>
</table>

*IC – intracranial; **Sp – Spinal

The age range for the group of intracranial resected meningiomas was 13 years to 80 years with an average age at resection of 45 years. There was a gradual increase with advancing age of the number of meningiomas resected with a peak in the 31 to 50 year age group (Table 2; Fig. 1). The male to female ratio for this group was 1.28 with no difference noted in the average age of resection for both sexes.

Figure 3

Spinal meningiomas accounted for 10% of the total number of meningiomas (Table 1). The age range was 19 years to 76 years (Table 2; Fig. 1) with an average age at resection of 55 years. The male to female ratio for this group was 1:7.

Of the seven meningioma cases described at autopsy, three were incidental findings. The remaining four had been diagnosed antemortem via imaging techniques; surgery had not yet been done to confirm the diagnosis for a variety of reasons. The age range for the autopsy meningioma group was 44 to 80 years (Table 2; Fig 1) with an average age of 55 years. All were intracranial in location and within the supratentorial compartment. No men were identified within this group.

No child under the age of 10 years was identified with a meningioma; however, three persons under the age of 20 years were identified (Table 2). One was a 19 year old woman with a spinal meningioma. The other two were both 13 years old; one was a boy with a frontal lobe fibrous meningioma (WHO Grade I) while the other was a girl with a frontal lobe malignant tumor (WHO Grade III). Follow up notes for these patients could not be located.

Analysis of the topographic distribution of these tumors revealed that 60 (90%) of the total number of meningiomas were intracranial in location while 7 (10%) were spinal. Of the former group, 7 (12%) were from the autopsy files (Table 1). Of the surgically resected intracranial group, 46 (87%) occurred within the supratentorial compartment, with 2 (4%) infratentorial and 2 (4%) spanning the two compartments. No definite location was given for 3 (5%) of the intracranial lesions (Table 3). The latter group included one case of multiple intracranial meningiomas in a 52 year old female patient who had been treated with radiotherapy for a posterior fossa tumor some thirty years previously.

Figure 4

<table>
<thead>
<tr>
<th>LOCATION</th>
<th>NUMBER</th>
<th>Percent (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Supratentorial</td>
<td>46</td>
<td>87</td>
</tr>
<tr>
<td>Infratentorial</td>
<td>2</td>
<td>4</td>
</tr>
<tr>
<td>Transientorial</td>
<td>2</td>
<td>4</td>
</tr>
<tr>
<td>None given</td>
<td>3</td>
<td>5</td>
</tr>
<tr>
<td>TOTAL</td>
<td>53</td>
<td>100</td>
</tr>
</tbody>
</table>

Of the supratentorial meningiomas, the fronto-parieto-temporal convexity of the brain was the most common location (67%), followed by sphenoid wing lesions (12%). Uncommon locations in this study included the cavernous sinus, optic nerve and olfactory nerve sheath (Table 4). Both
infratentorial lesions were attached to the falx cerebelli.

**Figure 5**

Table 4: Distribution of Supratentorial Meningiomas at the UHWI (1994-2003)

<table>
<thead>
<tr>
<th>LOCATION</th>
<th>NUMBER</th>
<th>PERCENT (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Fronto-temporo-parietal lobes</td>
<td>34</td>
<td>63</td>
</tr>
<tr>
<td>Sphenoid wing</td>
<td>6</td>
<td>11</td>
</tr>
<tr>
<td>Parasellar region</td>
<td>3</td>
<td>6</td>
</tr>
<tr>
<td>Parasagittal region</td>
<td>3</td>
<td>6</td>
</tr>
<tr>
<td>Occipital lobe</td>
<td>2</td>
<td>4</td>
</tr>
<tr>
<td>Cavernous sinus</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>Optic nerve</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>Olfactory nerve sheath</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>TOTAL</td>
<td>53</td>
<td>100</td>
</tr>
</tbody>
</table>

The majority of the resected intracranial meningiomas were WHO Grade I lesions (94%); of these, 28 (50%) were of the transitional subtype. There were also equal numbers of both the meningotheial and transitional subtypes (11% each), which were the second most common subtypes. Of the seven spinal meningiomas which were all WHO Grade I, 3 (43%) were psammomatous. The histologic subtypes of the meningiomas in the autopsy group could not be ascertained.

There were 4 (6%) WHO Grade III lesions all of which were intracranial in location: one papillary meningioma and 3 malignant lesions. One of the latter was seen in a 13 year old girl. Of the patients with the malignant subtype, the male: female ratio was 1:1.

There were a total of 7 (12%) recurrences in the group of resected meningiomas. All were intracranial in location. Of this group, 2 (29%) were designated histologically as malignant (WHO Grade III). Of the remaining 5 which were all designated as WHO Grade I tumors, only 1 had documented evidence of brain invasion histologically while in 2 cases, incomplete resection was documented by the clinician. The interval to recurrence for the two malignant meningiomas was one and eleven years; the shortest time to recurrence was within the incompletely resected group, where the interval was 6 months and one year respectively (Table 5).

**DISCUSSION**

In a study published in 1987 by Char et al, the incidence of intracranial meningiomas at the UHWI was 21.15%, which, despite being amongst the higher incidences, is comparable to those of many other series from all over the world (3). The latter averages an approximate incidence of about 20% except for Africa where the incidence is thought to be closer to 30%. The total number of intracranial meningiomas at the UHWI during the fifteen year study period (January 1970 to December 1984) was eighty eight, with a male: female ratio of 1:1.68. The number of spinal meningiomas was seven which accounted for 7.3% of the total number of meningiomas identified during that period.

In the present study series which is over a period of ten years, January 1994 to December 2003, the total number of intracranial meningiomas was sixty (60), with a male: female ratio of 1:2.8, which is higher than the previous local study but which mirrors the ratios quoted by several other authors (3, 4). Spinal meningiomas in this study accounted for approximately 10% of the total number of meningioma cases identified and this is comparable to figures quoted in the literature (3). Likewise, in this study, spinal tumors showed a marked preponderance in women with a male: female ratio
It has been suspected for many years that estrogens and progestins have a role in the pathogenesis and/or growth of meningiomas; however, to date, there has been no large long term study done to better delineate this role. The evidence has so far been circumstantial. There have been studies that suggest that breast cancer and meningiomas occur together more frequently while there have been others which counteract this claim (3, 4). Likewise, the claim that meningiomas grow faster during pregnancy has not yet been adequately investigated. What is so far accepted is that many meningiomas do show some amount of estrogen or progestin reactivity; however, the significance of this still remains unclear. It is a fact that this study revealed the expected increased incidence in women. Estrogen and progestin immunoreactivity, however, was not determined in the cases within this series.

Other factors thought to be associated with the pathogenesis of meningiomas include ionizing radiation, previous head trauma and genetic predisposition (1, 2). The latter may take the form of neurofibromatosis type 2 (NF 2), a disorder in which patients are prone to developing multiple meningiomas, meningiomas at a younger age and/or malignant subtypes. Over the last few decades, loss of a suppressor gene on the long arm of chromosome 22 has been isolated in as many as 80% of meningiomas analyzed (3, 7). In this study, three patients under the age of 20 years were identified, one of which had malignant histology; unfortunately, the notes and family history of all three could not be located. Interestingly, the only patient in this study to have been diagnosed with multiple intracranial meningiomas had been treated with radiotherapy for a posterior fossa tumor some thirty years previously.

This study also revealed the well known fact that the incidence of meningiomas increases with increasing age, and as seen in Fig. 1, the majority (55%) of resected intracranial meningiomas occurred in the 31 to 50 year age group. It is also interesting to note that the average age of resection of intracranial meningiomas was at least ten years less than the average age of resection of both spinal and autopsy diagnosed meningiomas. Most meningiomas discovered at autopsy are thought to be incidental and had therefore caused the patient little or no problems during life. It therefore follows that the age of diagnosis for these patients tends to be higher than patients whose meningiomas are surgically resected. It is interesting to note that of the seven patients whose meningiomas were histologically diagnosed for the first time at autopsy, three (43%) were completely incidental with no documented evidence of the patient complaining of symptoms antemortem. The post mortem notes of the remaining four patients revealed that a presumptive diagnosis had been made via imaging before death; however, the patients died from other causes before resection and therefore before a histologic diagnosis could have been rendered. The notes of only one of those four patients clearly stated that surgery had been offered but was refused.

The locations of the meningiomas in the present series are similar to those quoted in many other series including that by Char et al (8). In the latter series, supratentorial meningiomas were the majority with infratentorial meningiomas accounting for 4% of the total while in this more recent series, infratentorial lesions accounted for 3 % of the total (Table 3). Meningiomas occurring over the cerebral convexities, the falx, parasagittal and sphenoid wing tend to be the most common locations and this was reflected in the distribution of meningiomas in this study (Table 4). The location of these lesions is quite important as it is one of the factors that will help to determine prognosis as recurrence is a major consideration in lesions which are incompletely excised, even if the lesion has a benign histology. In this study, there were a total of 7 documented recurrences; of these, 5 (71%) were histologically benign (WHO Grade I). Of this subgroup, the clinicians stated in 2 cases that the lesions were incompletely resected – one within the frontal lobe and the other said to be transtentorial in location. It is important to note that these 2 cases have the shortest time to recurrence. It has been a well stated fact that the extent of resection is the most important factor underlying recurrence (6). Details of all the cases which recurred are shown in Table 5.

Meningiomas may also recur if they have the capacity to behave aggressively as with the atypical (World Health Organization, WHO Grade II) or malignant (WHO Grade III) sub-types. In this study, there were seven documented recurrences; of these, 2 (29%) were malignant (WHO Grade III) (Table 5). It is also very interesting to note that there were no atypical (WHO Grade II) tumors diagnosed during this study period. There are several criteria that were set out by the WHO in 1993 and revised in 1999 in Lyon, France (9) for a relatively accurate grading scheme; as such, none of the meningiomas in this study group fulfilled the criteria to be called Grade II. However, the WHO group has categorically
stated that even Grade I meningiomas which show brain invasion histologically will tend to have a clinical course similar to an atypical (Grade II) meningioma. It is therefore very unfortunate that usually no brain tissue is submitted at the time of resection for pathological assessment of brain invasion.

The most common subtype within the intracranial resected group was transitional (50%) followed by the fibrous and meningothelial subtypes (11%) which were present in equal numbers. These are all benign meningiomas (WHO Grade I) and it is noteworthy that malignant subtypes accounted for 6% of the total. This is similar to figures out of Manitoba, Canada where malignant meningiomas accounted for 7% of the total. This is similar to figures out of Bombay, India where malignant meningiomas accounted for only 1.9% of the total. It is also well known that malignant meningiomas are more common in men; in fact, in a large ten year study out of the Mayo Clinic, it was stated that the male gender was regarded as a negative prognostic factor. This is reflected in this study with the male to female ratio being 1:1 within this malignant sub-group as opposed to the overall ratio of 1:2.8.

In the group of spinal meningiomas, the psammomatous subtype was predominant (43%). This predominance has been noted by several other authors and one author has even suggested that patients with this subtype actually have a less favorable neurological outcome postoperatively. The reasons for the tendency to develop this subtype around the spinal cord are not fully understood.

It is therefore obvious from this retrospective study of meningiomas that their occurrence and behavior at this institution is similar to those at other institutions all over the world. This study also highlights the fact that the extent of surgical resection as well as the documentation of brain invasion histologically is of vital importance, as recurrences are quite likely and therefore careful patient follow up is indicated. It is hoped that in the near future enough information will be garnered which will enable adequate treatment plans for all patients with meningiomas, even those with sinister histologic subtypes and/or difficult to reach tumors.

References
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