



Research Paper

Trends in incidence of Ewing sarcoma of bone in India – Evidence from the National Cancer Registry Programme (1982–2011)



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ABSTRACT

Background: Ewing sarcoma is a malignant tumour found mainly in childhood and adolescence. The present study aims at analyzing the data on Ewing sarcoma cases of bone from the National Cancer Registry Programme, India to provide incidence, patterns, and trends in the Indian population.

Materials and Methods: The data of five Population Based Cancer Registries (PBCR) of Bangalore, Mumbai, Chennai, Bhopal and Delhi over 30 years period (1982–2011) were used to calculate the Age Specific and Age Standardized Incidence Rates (ASpR and ASIR), and trends in incidence was analyzed by linear and Joinpoint Regression.

Results: Ewing sarcoma comprised around 15 % of all bone malignancies. Sixty-eight percent were 0–19 years, with 1.6 times risk of tumour in bones of limbs as compared to other bones. The highest incidence rate (per million) was in the 10–14 years age group (male – 4.4, female – 2.9) with significantly increasing trend in ASpR observed in both sexes. Pooled ASIR per million for all ages was higher in male (1.6) than female (1.0) with an increasing rate ratio of ASIR with increase in age. Trend of pooled ASIR for all ages was significantly increased in both sexes. Twelve percent cases were reported in ≥ 30 years of age.

Conclusion: This paper has described population based measurements on burden and trends in incidence of skeletal Ewing in India. These may steer further research questions on the clinical and molecular epidemiology to explain factors associated with the increasing incidence of Ewing sarcoma bone observed in India.

1. Introduction

Ewing sarcoma is a malignant small round blue cell tumor of bone, first described by James Ewing as an endothelioma of bone in 1921 [1]. It is the third commonest primary malignant bone tumor in all age-groups following osteosarcoma and multiple myeloma. Among children and young adults, it is the second frequently occurring bone malignancy [2–4]. Studies have shown lower incidence of this tumour among the African and East Asian population as compared to the Whites [5,6] with a male preponderance [6,7]. Most studies in the Asian population are hospital or laboratory based and focussed on treatment outcomes with

no recent population based evidence of bone sarcoma incidence from India [8–12]. The Bombay Cancer Registry, the oldest population based registry in India, had reported Ewing sarcoma as the commonest bone malignancy in the late part of the 90s [13]. The Population based Cancer registry reports of the National Cancer Registry Programme, India provided incidence of all bone malignancies but description by specific histologic types was not presented [14]. This paper provides a detailed analysis of the magnitude of incidence and burden of Ewing sarcoma of bone in the Indian population based on thirty years' data from the Population based cancer registries (PBCR) of the National Cancer Registry Programme (NCRP), India.

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2. Material and methods

The Population based cancer registry data from five urban places – Bangalore (1982–2010), Mumbai and Chennai (1982–2011), Bhopal (1988–2011) and Delhi (1988–2009) have been used for the analysis. A total number of 8394 bone malignancies (C40 and C41 codes of the International Classification of Disease, 10th revision (ICD-10) [15] constituting approximately 1.2% of all cancers were registered in the NCRP. Among these, cases with morphology code M-92603/6 of the International Classification of Disease, Oncology – third edition (ICD-O-3) [16] were considered as Ewing sarcoma of bone. Peripheral neuroectodermal tumour (PNET) and Askin tumor also of the Ewing sarcoma family of tumours were not included as they had a different morphology code. Variables like age, sex, method of diagnosis, morphology, topography were extracted. Population estimations and quality checks on the data were based on the methods used by the NCRP, India [14].

Proportions of cancer type and site of cancer by age and sex groups were calculated. Unpaired t-test (continuous data) and odds ratio with 95% confidence limits (categorical variables) were calculated using SPSS version 22. Age Specific and Age Standardized (to world standard population) Incidence Rates (ASpR and ASIR) per million populations were calculated by age group and sex for each of the five PBCRs and the pooled data of five PBCRs. Standardized Rate Ratio of ASIR by sex group with its 95% confidence interval was also calculated.

Trends in ASpR and ASIR were calculated by five year periods from 1982 to 2011 using linear regression (IBM SPSS statistics for windows, version 22.0, Armonk, New York), and Joinpoint regression (Joinpoint Regression Program 4.0.1 January 2013, SEER) and expressed as Annual Percentage Change (APC) with significance at $p < 0.05$.

3. Results

Among all bone malignancies (8394) registered, 1301 (15.5%) cases were Ewing sarcoma of bone in the five PBCRs. In the childhood (0–14 years) group, Ewing sarcoma accounted for 577 (38.8%) of all bone malignancies in the PBCRs. The highest proportions were observed in 10–14 years age group (25.2%) followed by 15–19 years (23.3%), with 68% cases in the 0–19 years age group. The proportion of cases in ≥ 30 years age was 11.6% ($n = 150$) in PBCRs. The M:F ratio was 1.8:1. Microscopic diagnosis was available in 99.4% of all registered cases.

The Age Standardized Incidence Rate (ASIR) per million (pooled) for all ages were 1.6 (male) and 1.0 (female) with a Rate Ratio (RR) of 1.54. Pooled ASIR was the highest in 0 to 19 years (male – 2.8, female – 2.0) and the lowest in ≥ 30 years (male – 0.5, female – 0.2). Rate Ratio of ASIR was 2.46 in ≥ 30 years as compared to 20–29 years (1.65), and 0–19 years (1.41) (Table 1).

The Age Specific Incidence Rate (ASpR) per million varied from 0.5 (male) and 0.4 (female) in 0–4 years age group in Mumbai to 6.5 (male) and 4.4 (female) in the 10–14 years age group in Delhi. In all the

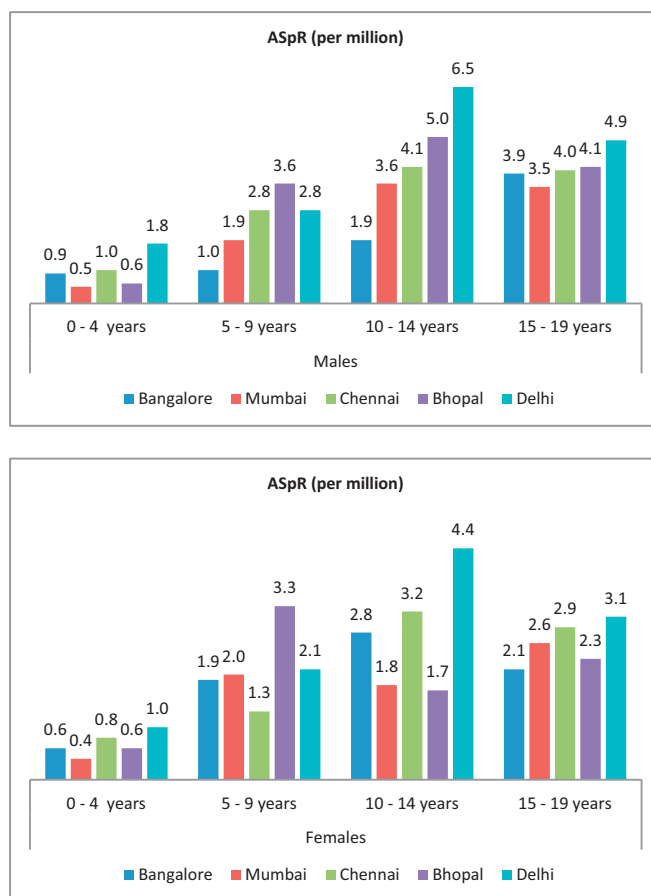


Fig. 1. Comparison of Age Specific Incidence Rate (ASpR) per million of Ewing's Sarcoma of bone by age group (0–19) and sex in five PBCRs (1982–2011), India.

PBCRs, the Age specific incidence rate peaked in the 10–14 years age group in both sexes except for females in Bhopal and Mumbai and males in Bangalore (Fig. 1).

The Age Specific Incidence Rate (ASpR) per million (pooled) was highest in the 10–14 years age group with 4.4 (male) and 2.9 (female) per million.

Pooled ASIR per million for all ages increased from 1.0 in 1982–86 to 2.0 in 2007–11, ($p = 0.019$) in males and from 0.8 to 1.2 ($p = 0.034$) in females with a five-year Annual Percentage Change (APC) of 2.24% (males) and 1.17% (females). A statistically significant increase in ASpR was observed in 10–14 years age group in both sex groups (Fig. 2).

Bones of extremities were more commonly involved compared to other bones (includes axial skeleton) in PBCRs (69.2%). The risk of

Table 1

Number, proportion (%), age–group specific (ASpR) and age standardized (ASIR) Incidence rate per million with Rate ratio and 95% Confidence Interval of Ewing Sarcoma of bone by sex in different age groups in five PBCRs (pooled) (1982–2011), India.

Age group (years)	Total n(%)	Male n(%)	Female n(%)	ASpR per million		ASIR per million		Rate ratio of ASIR (95% CI)
				Male	Female	Male	Female	
	1301 [100.0]	838 [64.4]	463 [35.6]					
00–04	70 (5.4)	44 (5.3)	26 (5.6)	1.1	0.7	2.8	2.0	1.41 (1.38–1.45)
05–09	181 (14.0)	99 (11.9)	82 (17.7)	2.2	2.0			
10–14	326 (25.2)	203 (24.4)	123 (26.6)	4.4	2.9			
15–19	302 (23.3)	196 (23.5)	106 (22.9)	4.1	2.7			
20–24	171 (13.2)	113 (13.6)	58 (12.6)	2.1	1.3	1.7	1.0	1.65 (1.53–1.78)
25–29	95 (7.3)	64 (7.7)	31 (6.7)	1.3	0.7			
> =30	150 (11.6)	114 (13.7)	36 (7.8)	0.6	0.2	0.5	0.2	2.46 (2.19–2.76)
All ages ^a	1295 (100.0)	833 (100.0)	462 (100.0)	1.7	1.1	1.6	1.0	1.54 (1.49–1.60)

^a Age not known for 6 cases.

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