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An Indian perspective on gnathic osteosarcoma: A comprehensive literature review of the last three decades

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ABSTRACT

Objective: In this review a meta-analysis of demographics, clinicopathologic features, histopathology, treatment aspects and future strategies of gnathic osteosarcoma (GOS) in Indian population was done and compared with the similar features in reported population based studies on GOS.
Study design: A computer aided English literature search was carried out by combing relevant search terms from the year 1981 till December 31st 2012. A total of 39 cases of gnathic osteosarcoma in Indian population from 27 published articles were compiled together and data regarding demographics, clinicopathologic features, histopathology, treatment aspects and future strategies were extracted.
Result: GOS happens to occur at a younger age group (3rd decade). We observed a slight male predominance and mandible (59%) being the most commonly involved jaw. An unusually wide latency period ranging from 14 days to 6 years has been noticed. Possible association of osteoblastic variant of osteosarcoma with negative prognosis (58%) may be a significant finding of this review. Juxtacortical gnathic osteosarcoma (18%) cannot be considered as so rare in Indian population.
Conclusion: Although advanced treatment modalities are followed in Indian population there were many lacunae while reporting cases. Immunohistochemistry, laboratory assay of alkaline phosphatase and lactate dehydrogenase and gene mapping of GOS patients should be part of the diagnostic protocol.

Contents

Abbreviations: GOS, gnathic osteosarcoma; MSCs, mesenchymal stem cells; ALP, alkaline phosphatase; LDH, lactate dehydrogenase.

☆ AsianAOMS: Asian Association of Oral and Maxillofacial Surgeons; ASOMP: Asian Society of Oral and Maxillofacial Pathology; JSOP: Japanese Society of Oral Pathology; JSOMS: Japanese Society of Oral and Maxillofacial Surgeons; JSOM: Japanese Society of Oral Medicine; JAMI: Japanese Academy of Maxillofacial Implants.

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1. Introduction

Osteosarcoma is a rare malignant bone neoplasm characterized by formation of disorganized immature osteoid tissue, mostly arising in the metaphyses of long bones [1]. Osteosarcoma of the jaw bones or gnathic osteosarcoma (GOS) accounts for nearly 6–13% of all osteosarcomas [2]. Incidentally GOS is more common in elderly population is less aggressive and has a tendency to spread locally than to produce distant metastases [3–5]. These findings may have led certain authors to suggest GOS to be a separate entity altogether, although considerable conflicts of opinion exist in this regard [3–5]. Due to population specific genetic influences there may be considerable differences in the incidence, demographics and clinicopathologic presentation of GOS depending upon the geographical location and ethnicity as reported by Beck et al. in Ewing's Sarcoma [6]. We have performed a meta-analysis of all the relevant characteristics of GOS in Indian population and compared with other population based studies, which will help the clinician to acquire in depth knowledge about GOS in Indian population. For literature review, a computer-aided literature search in Google Scholar was conducted by combing search terms "osteosarcoma", "Gnathic", "Jaw", "Maxilla", "Mandible" and "Bone sarcoma." The deadline of the included articles was December 31st 2012 starting from 1981. Reference list from the primary identified studies were also searched to prevent missing any studies by the electronic search strategies. We have summarized 39 cases from 27 articles of peer reviewed journals spanning 31 years from 1981 to 2012.

Only case reports of GOS in Indian population were included in this review. The cases of patients treated with prosthetic rehabilitation were excluded from the review assuming that they would have been reported as single case reports elsewhere. A data collection sheet was developed to record the following parameters like authors, year of publication, journal name, number of patients involved, sex, age, involved jaw, precursor lesion, clinical features, histological type, management and follow up. We have tabulated total 39 cases from 27 published articles [7–33] (Table 1) between the years 1981 and 2012.

Rare lesions like GOS should be studied in this retrospective way which provides data regarding incidence, prevalent age, sex, site, clinical presentation, histopathology type and treatment plan and follow up. Though there are many reviews of GOS available in the literature, there have been no reviews done in Indian population after Vege's review in 1981 [34]. Vege et al. has done an institutional study in 33 cases of craniofacial osteosarcoma between the years 1963 and 1981 over a 19-year period [34]. The present study will help to understand the transformation, if any, of the demographics, clinical presentation, awareness in the treatment protocol and other advanced concepts of GOS.

2. Pathogenesis

Most GOSs develop spontaneously without any obvious cause [35]. Reported risk factors of craniofacial osteosarcoma, are precursor lesions like osteo-dystrophia deformans [36], fibrous dysplasia [37], multiple osteochondromatosis, and myositis ossificans [38]. Radiation [39] and trauma [40] are reported to be very rare causes of GOS. It has been noted that radiation-induced osteosarcoma displays a higher aggressiveness with poorer prognosis than de novo-tumours [41]. The development of GOS in association with dental implant has been reported only once [42]. The most common site for the tumour is the rapidly growing endplate of the long bones which correlates with the fact that onset is usually during adolescent growth spurt. The fact that the appearance of GOS peaks 1 or 2 decades after adolescence might exclude growth as a major etiologic factor. Development of osteosarcoma is also associated with higher dose exposures of anthracyclines or alkylators and as second neoplasm in other malignancies like paediatric soft and bone tumours, renal tumours and Hodgkin's lymphoma [43]. Among our study population trauma and radiation were the possible risk factors in two patients. Synchronous presentation of GOS with extragnathic osteosarcoma in femur was reported in one of our cases and the presentation of GOS preceded trauma. In the above-mentioned case the author has excluded the possibility of metastatic osteosarcoma in mandible by the virtue of previous studies [44,45] and the presenting features of this patient. In this particular patient, whether trauma or genetic constitution of the patient or synergistic effect of the two risk factors is responsible for osteosarcomagenesis is not understood. Similarly we are not able to explain the pathogenesis for the patient who developed GOS after chemotherapeutically treated for Hodgkin's lymphoma. These kinds of patients should be sent for genetic mapping and there is no information about genetic mapping in any of our cases. This has reflected lack of awareness of genetic evaluation of osteosarcoma patients in Indian population.

Osteosarcoma is thought to originate from genetic and epigenetic alterations of multipotent mesenchymal stem cells (MSCs) or progenitors in the osteoblast lineage that retains the bipotency.

Table 1
List of authors and number of cases.

Number of cases	Name of author	Year of publication
1	Banerjee [7]	1981
3	Rao [8]	1983
8	Doval [9]	1997
1	Shah [10]	2000
1	Prakash [11]	2005
1	Balwani et al. [12]	2006
1	Kaveri [14]	2008
1	Dixit [13]	2008
1	Mohindra [15]	2009
1	Behere [16]	2009
3	Shetty [17]	2009
1	Basu [18]	2009
1	Desai [19]	2010
1	De [21]	2010
1	Sinha [20]	2010
1	Sethi [22]	2010
2	George [23]	2011
1	Verma [31]	2011
1	Narwal [24]	2011
1	Agarwal [25]	2011
1	Vaidya [26]	2011
1	Devi [27]	2011
1	Simon [28]	2011
1	Idiculla [29]	2011
1	Uma [30]	2011
1	Bojan [32]	2012
1	Sukumaran [33]	2012
39 patients	Total 27 articles	