Odontogenic Myxoma: A Rare Case of Aggressive Tumor with an Interesting Treatment Strategy

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ABSTRACT

Odontogenic myxoma is a relatively rare, benign odontogenic tumor of the oral cavity which is notorious for its inherent locally aggressive behaviour. There is currently no consensus on surgical management guidelines for odontogenic myxoma due to its rare incidence and a plethora of management techniques have been used. We report of a rare case of odontogenic myxoma in mandible and also discuss a novel approach wherein we did enucleation of the tumor followed by application of carnoy's solution as an adjunct which not only avoided radical resection of mandible and its associated co-morbidities but also prevented its recurrence which is the main reason for treatment failure. No recurrence of tumor has been seen over a period of three years.

KEYWORDS: Aggressive Tumor, Carnoys Solution, Odontogenic Myxoma, Odontogenic Tumors

INTRODUCTION

Odontogenic Myxoma of the jaw was first described by Thoma and Goldman (1947).In the International Histological Classification of odontogenic tumors, Odontogenic Myxoma (OM) is defined as a benign odontogenic tumor of mesenchymal origin that is locally invasive and consists of rounded and angular cells that lie in abundant mucoid stroma.[1] It is a tumor surrounded by Dogmas and Controversies.

Bryant (1802) introduced the term Myxosarcoma, which he described as a mucous transformation of round cell sarcoma. Myxomas were also described as Collenomas by Johannes Miller (1838). In his 1858 article titled Cellular Pathologie, Rudolph Virchow introduced the term Myxoma to describe soft tissue tumour resembling the structure of umbilical cord.[2] In 1948, Stout reported a series of 49 patients with myxoma and established criteria to be satisfied for diagnosis[2]—a true mesenchymal neoplasm consisting exclusively of undifferentiated stellate cells in a loose mucoid stroma that did not metastasize.[3]

CASE REPORT

A 26 year old female was referred to the department of oral and maxillofacial surgery by an orthodontist for the presence of a swelling over the left side of mandible since two months (fig. 1). The patient was undergoing orthodontic treatment for the past six months. Initially, the swelling was small in size and showed a gradual increase to attain the present size. Clinical examination

Fig No.1

revealed a firm, non-tender swelling expanding the buccal and lingual cortices of the mandible, extending from left first premolar region to third molar region, and it obliterated the buccal vestibule. The skin over the swelling was normal, and there was no history of paresthesia (fig.2). The panoramic radiograph showed a large, well-defined, multilocular radiolucent lesion with sclerotic margin and "soap bubble" appearance extending from the lower left second premolar to 5mm distal to the third molar (fig.3). The mandibular occlusal radiograph showed radiolucent lesion with expansions of buccal and lingual cortices(fig.4). Fine needle aspiration was performed to rule out odontogenic cysts, and results were negative. Considering the lesion to be benign odontogenic tumor, an incisional biopsy was made and a histopathological examination of the tissue sample revealed rounded, stellate, and spindle-shaped mesenchymal cells arranged in a loose, myxoid stroma with few collagen fibrils. These results were suggestive

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Fig No.2

Fig No.3

Fig No.4

of OM. Enucleation and curettage of the neoplasm were done along with the application of carnoy's solution (fig.5)under general anesthesia. The case is under follow up for 3yrs, with radiographs taken showing no evidence of recurrence (fig.6,7,8).

Fig No.5

Fig No.6

Fig No.7

Fig No.8

DISCUSSION

Odontogenic myxoma is regarded as a locally invasive tumour which does not metastasize and exhibits slow expansion, sometimes resulting in perforation of the cortical plates of the affected bone (Abiose *et al.*, 1987)[4]. Odontogenic myxomas are rare tumours and account for 3.3-15.7% of all odontogenic tumours in adults and for 8.5-11.6% amongst children. They occur in all age groups with a peak incidence in the third decade[5].The posterior region of the mandible are most frequently involved as is

seen in our case also. In many cases, these lesions are diagnosed accidentally by a routine dental check-up, and patients are mostly in their second or third decade of life [6] which is similar to our case. Odontogenic myxomas are thought to arise from the primitive mesenchymal structures of developing teeth which include dental papilla, follicle or periodontal ligament cells. Barros *et al.* argue that the tumour's embryologic origin from the tooth can be supported by the high frequency of this intraosseous tumour in the jaw with an extreme rarity in other bones of the skeleton.

Treatment Modalities And Prognosis: The tumour is not radiosensitive and consequently, has to be managed surgically. However, there has been some debate as to the best surgical approach. Surgical treatment of odontogenic myxoma vary from simple enucleation and curettage to segmental resection and hemimandibulectomy. Recurrence rates are reportedly high, around 25%, when a more conservative approach is taken.[7] Twenty of the 44 cases (45.5%) were managed by conservative surgical techniques, the remainder underwent radical surgery. Tumors recurred in three of those who were managed using conservative surgical approach(15.0%) There were no reported recurrences in any patient who underwent radical surgery.[8]

Reasons for recurrence: The lesions are not encapsulated thus allowing substantial infiltration into the adjacent medullary bone. There may be nests, pockets or loculations of the myxoid tumor which may hide behind bony trabeculations making it a stubborn infiltrative lesion. So the main reason or recurrence is thought to be incomplete removal rather than the intrinsic biological behaviour of the tumor.[9]

Treatment of these lesions, has a number of dilemmas about the choice and the degree of radicalism in surgical procedure that needs to be used, with the aim of decreasing the potential for recurrence, and minimizing the post-operative morbidity . In other words, the current controversies are leading to the crucial question: When and whether aggressive therapy is necessary in the treatments of this tumor. Conservative treatments have various advantages over segmental or block resection, and hemimandibulectomy with reconstructive surgery. Conservative treatments are less invasive, can be achieved by means of an intraoral surgical approach, preserve function and aesthetics, have a shorter hospitalization time, and are more cost-effective.[7]

According to Boffano *et al.* conservative treatment by enucleation and curettage is recommended when odontogenic myxoma is of less than 3 cm, whereas a segmental resection with immediate reconstruction is preferred in patients with larger tumors.[10] Recently, Zanetti *et al.* advocated that conservative treatment should involve enucleation of the lesion with a wide curettage of normal tissue or , or even peripheral osteotomy, as this has the advantage of preserving vital structures and also maintaining oral functional harmony.[11]

As in our case the tumor size was less than 3cm in its widest dimension on the radiograph, we used a conservative method of management which included enucleation of the tumor and using carnoys solution as an adjunctive.

The use of Carnoy's solution as an adjunct in the surgical management of Odontogenic Keratocyst and its correct application is of non questionable importance.[12] This technique of using carnoys as an adjunct lowers the recurrence rate (compared with enucleation alone) and morbidity (compared with resection) for aggressive and recurring tumors.[13] Carnoy's solution was first used as a medicament in surgery by Cutler and Zollinger in 1933. Carnoy's solution is prepared by mixing 3 ml of chloroform, 6 ml of absolute ethanol, 1 ml of glacial acetic acid and 1 g of ferric chloride.[14] This should be enough to do cauterization of the remaining cells. Its average depth of bone penetration depends upon the length of application (1.54 mm after 5 min).[15]

The effects of Carnoy's solution on the inferior alveolar nerve were first reported by Frerich. The authors did not observe axonal damage during the first three minutes of direct application.[16] In our case also there were no signs of inferior alveolar nerve paresthesia/anesthesia. So, in addition to enucleation or curettage the application of Carnoy's solution would be a better option for odontogenic myxoma. We would like to advocate that the application of Carnoy's solution is to preserve and save the unaffected bone so as to maintain the normal contour and architecture.

A follow- up period is clearly also necessary. It has been recommended that patients should be followed for at least the first two years after surgery during which the tumor has highest propensity for recurrence[4]. Our case was followed up for a period of three years with regular radiographs. No evidence of recurrence is seen clinically or radiographically. Moreover, we see a radiographic evidence of bone deposition in the defect (figure 7,8).

CONCLUSION

We would like to conclude that Carnoy's solution could be considered as a definitive and conservative management procedure in the treatment of these benign but aggressive lesions and would like to encourage maxillofacial surgeons to use this novel conservative method and document their follow-up data so as to formulate an evidence based treatment strategy. The successful surgical management of this case and its favourable prognosis should help the surgeons in taking treatment decisions for odontogenic myxoma, to minimize the risk of recurrence while adopting a less radical surgical approach whenever possible.

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