

A Rare Slow Growing Lump Pathosis - A Case Report

Case Report

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Abstract

Osteosarcoma is known for its slow growing nature. It is a rare pathology with characteristic lumpy pathosis of bones. It is mostly neglected/not noticed in the initial stages by the patient. In this case report, we present a case of a 23 years old female who was diagnosed initially to have aneurismal bone cyst or dentigerous cyst on clinical examination and cement-ossifying fibroma on a first in conclusive biopsy.

Patient presented with biopsy report done elsewhere which was in conclusive necessitating us to do a repeat biopsy. The biopsy report done in our institute gave a diagnosis of low grade osteosarcoma and in view of that we changed the treatment plan to a more aggressive approach. With this case report we would like to emphasize on a correct biopsy report to start treatment and also we discuss optimal management of head and neck osteosarcoma especially mandibular osteosarcoma.

Keywords: Osteosarcoma; Surgery; Low Grade; Swelling; Radiotherapy.

Introduction

The word "LUMP" means-mass which is indefinite in size and shape. 'Pathosis' defined as an abnormal diseased condition or a state.

Osteosarcoma is rare and slow growing in nature. It is more commonly found in long bones like femora, fibula. Osteosarcoma in head and neck region is a rare occurrence which affects 1:10,000,000 persons per year. 6%-8% of all head and neck tumors are osteosarcoma. Osteosarcoma occurs most commonly either during the teenage years or during later part of the adulthood [1].

Osteosarcoma is an aggressive tumor, with high recurrence rate and is more prone for metastasis. They tend to recur locally after treatment [2]. Evaluation and treatment plan of a patient with osteosarcoma is challenging. Histopathological diagnosis plays a

crucial role in the diagnosis of a jaw osteosarcoma as with any other tumor with equivocal signs and symptoms. The optimal treatment of a head and neck osteosarcoma still appears to be complete resection. The role of adjuvant therapy is ill-defined and depends on the histological grade [2].

Even a single case study on osteosarcoma has a significant impact on assessing its clinical nature and the prognosis in other patients because of its rare nature. Hence we report a case of osteosarcoma that reported to our institute and discuss the management protocol followed in our institute.

Case Presentation

A 24 year old female reported to the department of oral oncology with a swelling over the left side the mandible with history of 2 years duration. Swelling was small and soft in nature, which gradually increased in size to become bony hard in consistency

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later. Patient had reported to us due to her aesthetic concern only with no history of pain and paresthesia. She did not seem to have any relevant systemic abnormalities. Her personal history regarding substance abuse was negative.

On Local Examination

Extra-oral Examination

Gross facial asymmetry found on her left side of her jaw, with no deviation of jaw on opening and closing. A well-defined swelling of approximately 5x3cm in size extending.

Superiorly - approximately 3cm away from zygomatic arch.

Inferiorly - till inferior border of mandible.

Anteriorly - up to the corner of mouth and

Posteriorly - approximately 2cm away from angle of the mandible.

The swelling was non-tender, non-fluctuant, with no associated paresthesia.

Intra-oral Examination

Swelling of about 4*2cm was evident at the left lower buccal vestibule region. It was bony hard in consistency, non-tender, non-fluctuant with the following extensions.

Anteriorly- upto the mesial surface of 33.

Posteriorly - up to the distal surface of 37.

Medially -confined to the lingual gingiva of 34 to 37.

On general medical examination, she gave a past surgical history

of undergoing c-section one and half years back without any complications. On reviewing the biopsy report from the previous institution, it was found that the report was inconclusive and hence a re-biopsy was essential to rule out low-grade osteosarcoma. Re-biopsy done at our institute confirmed the diagnosis of low-grade osteosarcoma (Figure1).

After confirmation of the diagnosis as low grade osteosarcoma following second biopsy, we had extended our investigation to PET CT to rule out the metastatic spread. Figures 2, show the imaging sections.

The report from PET-CT read as follows-Metabolically active disease is seen in the expansile destructive lytic lesion with intra-osseous soft tissue component in left mandible. Nodes noted on bilateral neck involving submandibular region, level IIA & jugular group of largest measuring 12.2 x 9.4mm in right level IIA & 11 x 9.5mm in left level IIA showing increased metabolic activity.

Surgical Treatment Plan - Wide local excision of the lesion + segmental mandibulectomy + Free fibula microvascular flap reconstruction of the left mandible under GA. Figures 3.

After two weeks of surgery patient was referred for CT and RT. Patient was also under monthly review and was monitored closely.

As per patient's wish, and also considering her age we opted for free fibula as mode of reconstruction which is also suitable for rehabilitation with implants later. We had made a good satisfactory follow-up close to two & half years (Figure 4).

Figure 1. biopsy report.

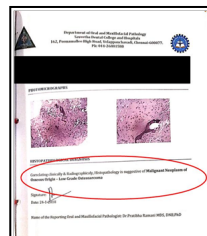


Figure 2. Coronal imaging section.

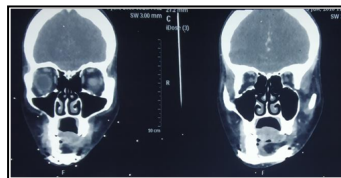


Figure 3. a-surgical markings. b-Skin Flap elevation exposing lesion. c-specimen. d-Fibula flap harvest. e-Fibula flap reconstructing the mandible. f-Post op OPG.



Figure 4. a-Pre-operative profile. b-Post-operative profile.

Discussion

Osteosarcoma is an aggressive form of bone malignancy characterized by osteoid formation by highly malignant osteoblasts [3] which is more commonly found in long bones followed by jaw bones (6% to 10%) and long bone osteosarcomas are more common in childhood-adolescent age groups [3]. Head and neck osteosarcomas are common in the fourth decade of life [2]. The male to female ratio is approximately 1:1 [2].

Another main difference between extremity osteosarcoma and head and neck osteosarcoma is in their pattern of metastasis. Head and neck osteosarcoma have a tendency for local recurrence [2] whereas osteosarcoma of the extremities have propensity for distant metastases. When you compare survival rates among maxillary and mandibular tumors, patients with mandibular tumors have greater survival potential. One reason could be the highly vascular nature of the maxilla and increased chances of metastasis due to this [3]. Other factors that influenced survival were histological grade and subtype, surgical margins. The three grades of osteosarcoma classified according to clinical nature are low-grade (para-osteal or central) intermediate grade (periosteal) and high grade (conventional, telangiectatic and small cell) [4].

In osteosarcoma of the extremities, adjuvant therapy improved overall survival. But the same could not be extrapolated to head and neck osteosarcomas because of low numbers and a retrospective cohort study was done by YiMing Chen et al in tertiary hospital to find out about the role of adjuvant therapy in head and neck osteosarcomas. From the study it was proven that adjuvant chemotherapy improved overall survival and adjuvant radiotherapy improved local control [3]. Hence, we discussed with our in house radiotherapist and medical oncologist and according to his suggestion we advised adjuvant radiotherapy and chemotherapy for the patient.

Smith et al., in another study summarise that there is similar 5 year survival rate between surgery only and surgery with chemotherapy even when poor disease characteristics like high grade and metastatic nature is taken in to account but they found that for determining overall survival chemotherapy played a significant role [3]. Adjuvant postoperative RT is also indicated for those patients with close or positive margins [2].

It is mandatory to come-up with multiple diagnostic aids such as CT, CECT, PET (more reliable for metastatic lesion like osteosarcoma) to rule out other bony lesions such as Paget's disease and others. The imaging report has to be correlated with clinical history and biopsy for confirmatory diagnosis. Dynamic MRI and PET are promising to assess tumor response (in addition to CT scan

and conventional MRI) and should be investigated further [4].

In our patient since the first biopsy was did not conclusively diagnose the lesion as cement-ossifying fibroma, we had to do second biopsy to conclusively diagnose the lesion and treat the patient. In our case doing a second biopsy turned out to be crucial because, according to the first biopsy the diagnosis was a benign lesion and it would have required only a conservative treatment approach and low grade osteosarcoma is treated aggressively for its metastatic nature and recurrence potential. The main treatment modality for head and neck osteosarcoma is complete resection with large bone and soft tissue margins. The decision to give adjuvant therapy varies among different institutions. The main deciding factor is the capacity to yield negative margin intra-operatively. The other factors that influence being risk of local recurrence and the possibility to functionally rehabilitate the patient [4].

To aid the surgeon in getting negative margins preoperative investigations play a major role. Intramedullary involvement should be assessed preoperatively and MRI plays a significant role in this. Intra-operative frozen section cannot determine marrow involvement [4].

For segmental resections free bone flaps like fibula is the preferred treatment option as the associated success rate is more and huge resections can be done with good cosmetic results. The options available to the surgeon are immediate reconstruction vs delayed reconstruction. Immediate reconstruction is suitable for young patients as delayed reconstruction in retracted and fibrotic soft tissues poorer results. The long-term functional, esthetic and psychosocial benefits are better with immediate reconstruction. We chose fibula in this patient with the aim of giving guided implants at a later date [5].

Following surgery, patient was referred for chemotherapy and radiotherapy. Adjuvant chemotherapy and radiotherapy improves local control in head and neck region [3]. Multiple studies have proven that surgery, chemotherapy and radiotherapy proved relevant in the treatment of head and neck osteosarcoma. Many studies suggested the main indication of postoperative radiotherapy indication is positive margin [5]. In our case we had referred the patient for post-operative radiotherapy and chemotherapy after discussion with our in house radiotherapists even though we did not have positive margin during surgery.

Patient had regular routine review and follow-ups for 2 and half years with disease free survival. We also took PET CT a month ago to rule out metastasis and recurrence. We are absolutely satisfied with the treatment outcome of the patient.

Conclusion

Rarest metastatic lesions like Osteosarcoma must be carefully examined to rule out even minimal chance of error even if it involves using multiple imaging techniques. We recommend MRI whole body or PET CT which is more reliable one with these kinds of metastatic lesions. It's mandatory for a second biopsy when there is a confounding first result as it is better to err on the side of caution.

Osteosarcoma requires aggressive treatment for minimizing the chances of recurrence and distant metastasis. Ultimate aim of the treatment should be, producing the overall good disease free survival.

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