Case Report

Giant cell tumor of cervicothoracic region treated by triple corpectomy from posterior only approach: A case report with review of literature

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INTRODUCTION

Jaffe, Lichstein, and Portis first defined the criteria for diagnosing GCT in 1940 to differentiate it from other giant cell containing lytic lesions of bones.[3] Giant cell tumor (GCT) of the spine is a rare entity and has been known as a benign aggressive tumor in the literature. It has been known to affect mobile as well as rigid and immobile segments of spine. The incidence of GCT is estimated to be 1.4-9.4% in the mobile segment of spine.[2] It affects females slightly more than males and affects people in their most productive years in a third or fourth decade. Metastases to lungs occur in approximately 3% of patients of GCT of bones.[3] It usually affects the vertebral body, but can affect posterior elements as well. Treatment of GCT has been variable with good results being shown by intralesional excision, en bloc spondylectomy, recurrent embolization, and also medical management with denosumab and bisphosphonates. We report a case of GCT involving three vertebral bodies C7, D1, and D2 at cervicothoracic region who presented to us and was treated with triple corpectomy from the posterior only approach. This is the first ever case report of triple corpectomy and anterior reconstruction by a posterior only approach for GCT at the cervicothoracic junction to the best of author’s knowledge.

Key words: Cervicothoracic, giant cell tumor, posterior approach, triple corpectomy
based corpectomy and anterior reconstruction from posterior approach itself followed by radiotherapy.

**CASE REPORT**

A 38-year-old lady presented with complaints of pain over the neck radiating to left upper limb for last 4 years with progressively increasing weakness in her left hand for last 2 years. She also complained of numbness in the left hand, left axilla, and on left side of chest. There was significant wasting of intrinsic muscles of the hand. On clinical examination, motor strength of her left long flexors and intrinsic muscles of hand were weak (2/5 as per MRC). There was hypoesthesia over C7, C8, and D1, D2 dermatomes. There were no long tract signs suggestive of myelopathy. Imaging studies were done which showed a lobulated tumor arising from C7, D1, and D2 compressing the left C7, C8, D1, and D2 roots [Figure 1]. Biopsy of tumor suggested GCT. The patient was operated from a single posterior approach. Preoperative tumor embolization was done to decrease intraoperative blood loss. Intraoperative neuromonitoring was used to detect deficit due to the handling of cord and nerve roots. Posterior stabilization using lateral mass screws in C4, C5, and C6 and pedicle screws in D3, D4, and D5 was done. Laminectomy extending from C6 to D2 was done, and a tapered rod was applied on one side. Facetectomies were done on bilateral sides, and roots were isolated and separated from the tumor mass. Roots were freed to the most far lateral extent possible to allow a greater space for insertion of cage among roots [Figure 2]. It also allows roots to be free and allows for some manipulation, which can occur while inserting cage. Corpectomies of C7, D1, and D2 were done from a posterior approach. Burr was used to decorticate the remaining anterior shell of bone from a posterior approach. Roots were slightly retracted under neuromonitoring to allow insertion of the cage from a posterior approach and anterior reconstruction with mesh titanium cage was done from a posterior approach. No root was sacrificed during surgery. D2 root was also preserved as sometimes intrinsic muscles of hand get a partial nerve supply from D2 if it is a postfixed plexus. It is difficult to identify preoperatively the exact motor supply of D2 root. Hence, it was decided to preserve all roots. The patient was put on radiotherapy postsurgery and was started on bisphosphonates. Patient recovered well and had improved grip strength 1-year postsurgery [Figures 3 and 4]. There was no recurrence of tumor at last follow-up.

**DISCUSSION**

GCT frequently affects epiphysis of extremities in a young population. It affects spine infrequently and affects sacrum more commonly than mobile regions of spine. It is a slow growing tumor and can persist as a mass long before it gets diagnosed. Common presenting complaints are pain over the affected site, weakness, and paresthesias due to compression of nerve roots. Eckardt and Grogan have recommended intrallesional curettage with adjuvant therapy for Stage I and II enneking lesions and en bloc should be reserved for most stage III lesions. However, this recommendation was for GCT involving long extremities.
Moreover, surgeon should remember GCT is primarily a benign lesion to begin with which can behave aggressively sometimes. *En bloc* spondylectomy is reserved for tumors, which involve a part of vertebral body and do not involve at least one pedicle. *En bloc* spondylectomy can be done at cervical region but requires careful isolation of vertebral arteries, which involves combined anterior, posterior approach, and can be a prodigious task at cervicothoracic region and may not be technically feasible in hands of all oncosurgeons. Literature also does not show clearly the superiority of *en bloc* resection over intraläsional resection and adjuvant treatment in these difficult to assess regions [Table 1]. Multiple authors have reported conservative resection in GCT of spine to give acceptable results. Guo et al. demonstrated acceptable recurrence rates and good clinical outcome after intraläsional resection of GCT in sacrum.[10] Junming et al. also suggested that *en bloc* spondylectomy may not be feasible in the cervical spine and intraläsional resection followed by radiation gives acceptable results.[7] Hart et al. reported that *en bloc* removal was more difficult to achieve and recurrence higher if the tumor extended into the posterior elements or into soft tissues.[4] Sanjay et al. reported *en bloc* resection in 10 patients and intraläsional resection in 14 patients with GCTs in mobile spine.[6] 50% of the patients with *en bloc* resection had a recurrence while 38% of those who had intraläsional resection had a recurrence. Authors suggested intraläsional resection as a minimum treatment modality for the treatment of GCT. In this case, tumor had already breached capsule and was compressing nerve roots of C7, T1-weighted, and T2-weighted on left side. Hence, *en bloc* spondylectomy was not a favored approach. Gross total resection was done with an intraläsional margin from posterior approach and patient was started on postoperative radiotherapy. Patient has been kept at regular follow-up and has been recurrence free at 1-year follow-up. This is a very short follow-up, as GCT has been known to recur as late as 60 months after index surgery.[2] The aim of the case report is to emphasize the technique and feasibility of doing triple corpectomy with anterior stabilization from the posterior only approach at the not so easily accessible cervicothoracic region. GCT has got a depraved standing of recurrence after surgery, and high recurrence has been reported in clinical series. Adjuvant treatment in the form of cryotherapy, phenol, recurrent embolization, and radiation has been described in the literature. There exists around ten percent risk of conversion of GCT to sarcoma after radiation, but potential benefits of radiation have also been described in preventing recurrence after intraläsional excision of GCT.[9] Combined anterior and posterior approach have been described in the literature for removal of the tumor but these approaches are morbid, time-consuming. Anterior approach at the cervicothoracic junction is difficult and requires sternal splitting approach and requires mobilization of great vessels. There is a lot less literature about management of GCT at cervicothoracic regions and no report involving three or more vertebra at this location to the best of our knowledge. Gille et al. reported a case of triple cervical corpectomy, which required ligation of bilateral vertebral arteries, and was done from combined anterior, posterior approach.[10] Bilateral vertebral artery ligation may not be possible in all patients and requires combined approach. Intraläsional removal of the tumor can be done at these

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### Table 1: Select series from literature emphasizing the treatment modalities used for tumors at cervicothoracic region

| Authors     | Number of patients | C7 | D1 | D2 | Surgery done | Recurrence | Adjuvant therapy |
|-------------|--------------------|----|----|----|--------------|------------|-----------------|
| Boriani et al. | 49 cases          | 1  | I  |    | IL excision  | Yes        | Not used        |
|             |                   | 1  | I  |    | IL excision  | No         | Radiotherapy    |
| Bhupendra et al. | 24 cases      |    |    | I  | IL excision  | No         | Not used        |
| Junming et al. | 21 cases         | C6-T1 |    | C6-C7 | Removal of C7 tumor | Yes        | Not used        |
|             |                   | C7 |    |    | Total resection | No    | RT              |
|             |                   | C7 |    |    | Total resection | No    | RT              |
|             |                   | C7 |    |    | Subtotal resection | Yes   | RT              |
| Gille et al. | Case report       | C5-C7 recurrent | C5-C7 vertebrectomy | No | RT |
| Rodrigues   | Case report       | C7 |    |    | En-bloc resection | No     | No              |

**Table 1**: Select series from literature emphasizing the treatment modalities used for tumors at cervicothoracic region.

IL: Intraläsional, RT: Radiation therapy
difficult to resect regions, but it leaves a definite risk of recurrence of GCT, which needs to be addressed with radiation or other adjuvant treatment and careful follow-up. Posterior-only approach allows for complete tumor removal along with circumferential stabilization in a single setting. There are numerous case reports, which also suggest the role of recurrent embolization, as solo treatment of GCT, and also role of agents such as denosumab and bisphosphonates are being explored.\cite{11,12} However, it is difficult to predict their role esp. when the patient has already developed neural deficit because of compression of nerve roots.

CONCLUSION

Aggressive en bloc resection of the tumor though gold standard might not be possible in all circumstances. Intralesional resection with adjuvant therapy from posterior only approach may be a suitable alternative for GCT involving multiple vertebra at the cervicothoracic region. Radiation is a double edge sword in GCT, and its pros and cons should be discussed with the patient. With the stereotactic radiosurgery available now, it might be possible to decrease the hazards of radiation to a minimum while piling the maximal gains.

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Conflicts of interest
There are no conflicts of interest.

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