Clinical Article

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Primary Osseous Tumors of the Subaxial Cervical Spine, Clinical Case Series

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Abstract

Background Data: Primary osseous tumors of the subaxial cervical spine are rare and can present with different clinical presentations and pathologies.
Study Design: Retrospective descriptive clinical case series.
Purpose: To assess the prognosis, surgical management, outcome of primary osseous tumors of the subaxial cervical spine
Patients and Methods: Eight patients underwent surgical treatment for primary cervical bony tumors at Kasr Al-Aini Medical School, Cairo University, between 2007 and 2012. Mean follow-up was 24.4±6.8 months. Clinical outcome were evaluated neurologically using Frankel grading system, while visual analog scale (VAS) has been used to evaluate neck pain.
Results: There was equal sex distribution and the mean age was 38.4±14.9 years (range 17-62 years). Half of patients had benign tumors (2 aneurysmal bone cysts, one hemangioma and one osteoblastoma) while the other 4 patients had malignant tumors (giant cell tumor, chordoma, chondro-sarcoma and plasmacytoma). Neck pain was the constant complaint of all patients. In addition, 5 patients suffered from myelopathy while 2 patients had radiculopathy. Anterior corpectomy and fusion was performed in four cases, while in 2 patients, only posterior approach was adopted. In the other 2 patients, combined anterior and posterior approaches were adopted. Postoperatively, 6 patients had the same preoperative Frankel grade (4 were grade E, 1 was D and 1 was C), while 2 patients improved (25%) (1 improved from D to E and 1 from C to D). The average VAS improved from 5±1.5 (range 3-8) preoperatively to 2.4±1.7 (range 1-5) postoperatively at last follow-up. The 4 patients with benign tumors are doing well (Frankel grade E). In contrast, 3 out of 4 patients with malignant tumors died within 12-16 m later, the survival of malignant tumors is 25% at 3 years.
Conclusion: Marginal piecemeal resection and cervical instrumentation and fusion is a safe and effective method in management of benign osseous cervical tumors. However, the results are poor for malignant tumors even with adjuvant therapy. (2013ESJ051)
Key words: cervical spine tumors, spinal neoplasm, cervical fusion

Introduction

Primary osseous tumors of the subaxial cervical spine are rare, representing less than 5% of all primary spine tumors of bone origin.[4,7,15,16] Because of their location, both neural and vascular structures can be affected.[14,20] The diagnosis of primary cervical tumors should be excluded in patients with persistent neck pain, brachialgia or quadriparesis, because of the varying clinical presentations and potential morbidity and mortality associated with these lesions.[4,6,8]

The pathology of primary osseous spinal tumors includes a different variety of benign and malignant neoplasms. Such tumors include aneurysmal bone cysts, hemangiomas, eosinophilic granulomas, osteoid osteomas, and osteoblastomas; chordomas, giant cell tumors, osteosarcomas, chondrosarcomas, synovial sarcomas. Tumors of the lymphoreticular system such as plasmocytoma, are generally considered in the discussion of spine tumors.[4,5,7,15]

The surgical excision of primary cervical bony tumors is challenging due to complex regional anatomy and the rarity of such lesions. So, the management of each neoplasm is often individualized on a case-by case basis.[10,11,14,20] Recently, the value of multidisciplinary approach for management of primary cervical tumors, particularly in cases of malignancy, cannot be overestimated.[1] The team approach to medical and surgical treatment enables the formulation of a coordinated perioperative care plan among treating physicians and surgeons of multiple specialties, which may include musculoskeletal oncology, radiotherapy, vascular surgery, otolaryngology and plastic and reconstructive surgery.[20]

This study represents a preliminary experience with the management of primary osseous neoplasms of the cervical spine and the various approaches adopted for tumor excision and spinal fusion with instrumentation.

Patients and Methods

This was a retrospective study conducted at Al-Manial university hospital, Kasr Al-Aini Medical School, Cairo University, between January 2007 and December 2012. Eight patients with primary subaxial cervical spine tumors of osseous origin were recruited. Patients with metastases or non-osseous origin were excluded from this study. Each case was analyzed according to sex, age, clinical presentation, radiographic findings, operative details, pathology and outcome.

Our patients were followed at outpatient clinic, where they were assessed clinically by history and physical examination, and neurologically by Frankel grading.[12] Pain has been assessed using the visual analog scale (VAS)[17] (0=no pain, while 10=the worst imaginable pain). All patients were radiologically evaluated by plain radiography, computed tomography (MS-CT) and magnetic resonance imaging (MRI) of the cervical spine. It is worth mentioning that all 8 cases had initial postoperative MRI and CT of the cervical spine to assess the cord decompression, extent of tumor excision and the sagittal alignment. At the follow-up, patients were evaluated with plain x-ray and if there was any significant complaint, follow-up CT was performed. The mean follow-up duration was 24.4±6.8 months (ranging 15-36 months). Table 1 shows the characteristics of the patients in this study.

Data were statistically described in terms of mean±standard deviation (SD) and range, or frequencies and percentages when appropriate. Comparison of VAS between pre and post-operative values was done using Wilcoxon signed rank test for paired (matched) samples. For comparing Frankel score, McNemar test was used. P values less than 0.05 was considered statistically significant. All statistical calculations were done using computer program SPSS (Statistical Package for the Social Science; SPSS Inc., Chicago, IL, USA) release 15 for Microsoft Windows (2006).

Results

There was no specific gender predilection (M/F=1/1). The mean age was 38.4±14.9 years (range; 17-62 years). Half of patients were classified as having benign tumors (mean age; 29.3±13.1 years) and the other 4 patients as having malignant tumors (mean age; 47.5±11 years). The pathology of the 4 benign tumors was 2 patients with aneurysmal bone cysts, one with hemangioma and one with osteoblastoma while the pathology of the other 4 malignant tumors was giant cell tumor, chordoma, chondro-sarcoma and plasmacytoma.

Neck pain was the constant complaint of all

patients. The average preoperative VAS was 5±1.5 (range; 3-8), which improved postoperatively (at last follow-up) to 2.41.7± (range; 1-5) which was found to be statistically significant (P=0.033). In addition, 5 patients suffered from myelopathy while 2 patients had radiculopathy. At time of presentation, 2 patients were Frankel grade C, 2 were grade D, and the other 4 cases were neurologically intact (Frankel grade E). Diagnostic evaluation of all patients included plain radiography, computed tomography (CT) and magnetic resonance imaging (MRI).

Tumors involved only the anterior part of the spine (vertebral body) in 5 cases and the posterior part of the spine (pedicle, facet and/or lamina) in 2 cases (Table 1). Both the anterior and posterior vertebral components were involved in one case. Treatment was individualized for each patient based on the site and type of the tumor (based on the preoperative imaging). Three patients underwent preoperative embolization of the tumors with polyvinyl alcohol to decrease the vascularity of the lesions.

Anterior corpectomy and fusion was performed in four cases when the tumor did not extend to the pedicles. While in 2 patients, only posterior approach was adopted. Posterior decompression alone was performed in one patient when the laminae were only involved without facet affection; while the other patient necessitated posterior decompression and lateral mass fixation due to involvement of the facets bilaterally. In the other 2 patients, combined anterior and posterior approaches were adopted because the tumor extended to both vertebral body and the posterior vertebral components. In one patient anterior corpectomy was associated with posterior decompression only; while in the other patient, posterior decompression was followed by lateral mass fixation. It is worth noting that 5 out of 6 patients who had anterior corpectomy underwent fusion via bone graft and plating while one patient with aneurismal bone cyst underwent fusion using bone cement only without plating. Figure 1-4 are examples of our patients' management.

In 6 patients (the 4 benign tumors and 2 malignant tumors: plasmacytoma and giant cell tumor), gross total excision was achieved via intralesional piecemeal resection (rather than margin-free en bloc resection because intraoperative pathological assessment was unavailable). It is worth noting that only de-bulking was feasible in 2 malignant tumors (50% of malignant tumors) due to infiltration of vital neuro-vascular structures; one case of chordoma which extended to the epidural space and infiltrated the vertebral and carotid arteries and one case of chondrosarcoma infiltrated the esophagus and carotid sheath.

Immediate postoperatively, 6 patients had the same preoperative Frankel grade (4 were grade E, 1 one was D and 1 was C). While 2 patients improved (25%) (1 improved from D to E and 1 from C to D). At the last follow-up, the patients had the same Frankel grading without deterioration. The difference between pre/postoperative Frankel grade was not statistically significant (P=0.37).

Regarding the benign tumors, one patient with an aneurysmal bone cyst had a recurrence after excision and underwent another operation without recurrence. The 4 patients with benign tumors are doing well (Frankel grade E) at last follow up. In contrast, out of 4 patients with malignant tumors, 2 patients had residual tumor (chordoma and chondro-sarcoma) and died within 12-16 m later. Regarding the other 2 patients who underwent gross total tumor excision; the patient with plasmacytoma did not suffer from local recurrence however he died 2y postoperatively from progression to multiple myeloma, while the patient with giant cell tumor had local recurrence and was reoperated and still doing well after 36 m follow up. It is worth mentioning that only the 4 patients with malignant tumors underwent radiotherapy postoperatively.

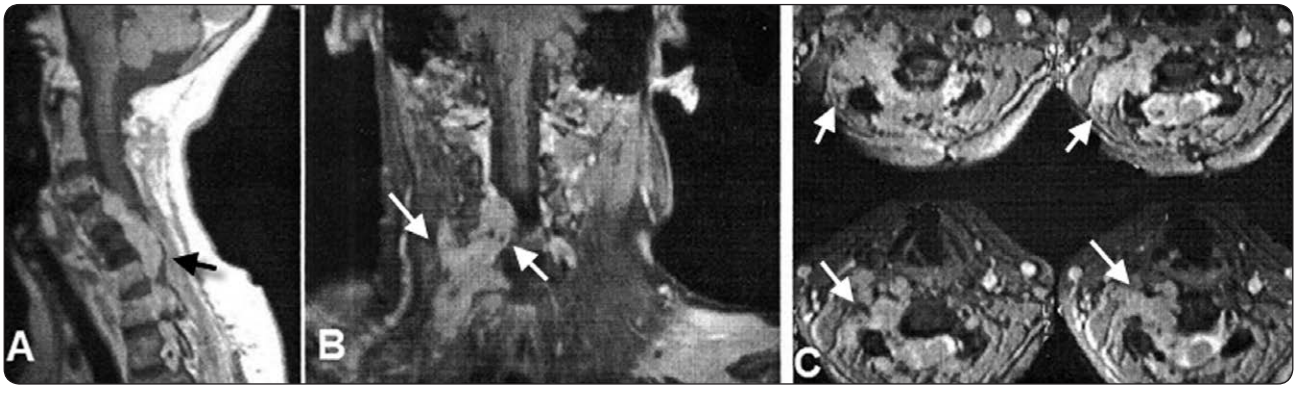
Solid fusion was evident in the follow up plain radiographs and CT of the cervical spine of 6 patients (the 4 benign tumors and 2 malignant tumors; giant cell tumor and patient with plasmacytoma who died 2 years postoperatively). Although the other 2 patients with chondrosarcoma and chordoma did not have clear evidence of bony fusion, they did not complain of instability before death.

Table 1: Data of Patients with Primary Osseous Cervical Tumors.

.No	Sex	Age	Presentation	Location	Level	Pathology	Surgery	Frankel grade		VAS		Outcome
								Pre	post	pre	post	
1	M	20	Neck pain, radiculopathy	Posterior	C4	Aneurysmal bone cyst	PD	E	E	5	2	Recurrence, reoperation
2	F	45	Neck pain	Anterior	C7	Hemangioma	ACF	E	E	4	1	No recurrence
3	M	17	Neck pain, myelopathy	Anterior	C6	Aneurysmal bone cyst	ACF	D	E	6	1	No recurrence
4	F	35	Neck pain, radiculopathy	Posterior	C5	Osteoblastoma	PDF	E	E	8	1	No recurrence
5	F	38	Neck pain, radiculopathy	Anterior	C5	Giant cell tumor	ACF	E	E	5	2	Recurrence reoperation
6	M	50	Neck pain & myelopathy	Anterior & Posterior	C6	Plasmacytoma	ACF& PF	D	D	4	2	Death 24m
7	M	40	Neck pain, myelopathy	Anterior	C4- 6	chordoma	ACF	C	C	3	5	Residual, Death 16m
8	F	60	Neck pain, myelopathy	Anterior & Posterior	C6	chondrosar-coma	ACF& PD	C	D	5	5	Residual, Death 12m

(ACF; anterior corpectomy and fusion, PD; posterior decompression, PDF; posterior decompression and fusion, Pre; preoperative, post; last postoperative follow-up, VAS; visual analogue scale)

Figure 1. (A) Sagittal (B) coronal magnetic resonance (MR) image showing the huge mass of a recurrent chordoma extending extradurally compressing the spinal cord from C4 to C6 (C) Axial MR images of the same patient showing the tumor compressing the spinal cord and involved with both the caroid and vertebral arteries (Note the arrows referring to the chordoma).



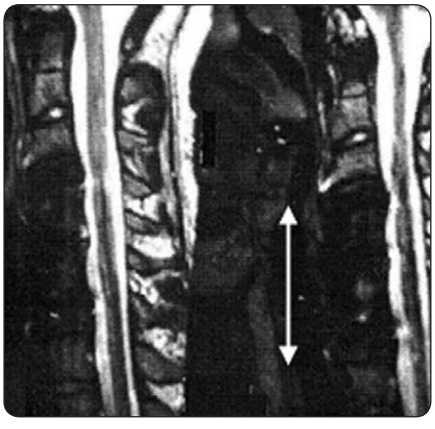


Figure 2. Postoperative MRI of the same patient showing debulking of tumor and corpectomy from C4 to C6, and fusion from C3 to C7 with a bone graft and plate.

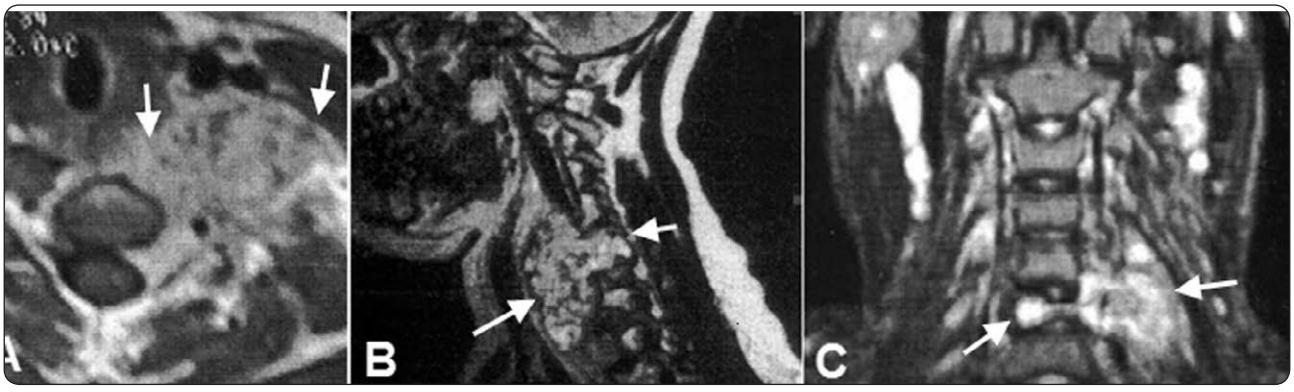


Figure 3. (A) Axial, (B) Coronal and (C) Sagittal MRI image showing the huge mass of aneurysmal bone cyst compressing the spinal cord at C6 (Note the arrows referring to the margins of the cyst).

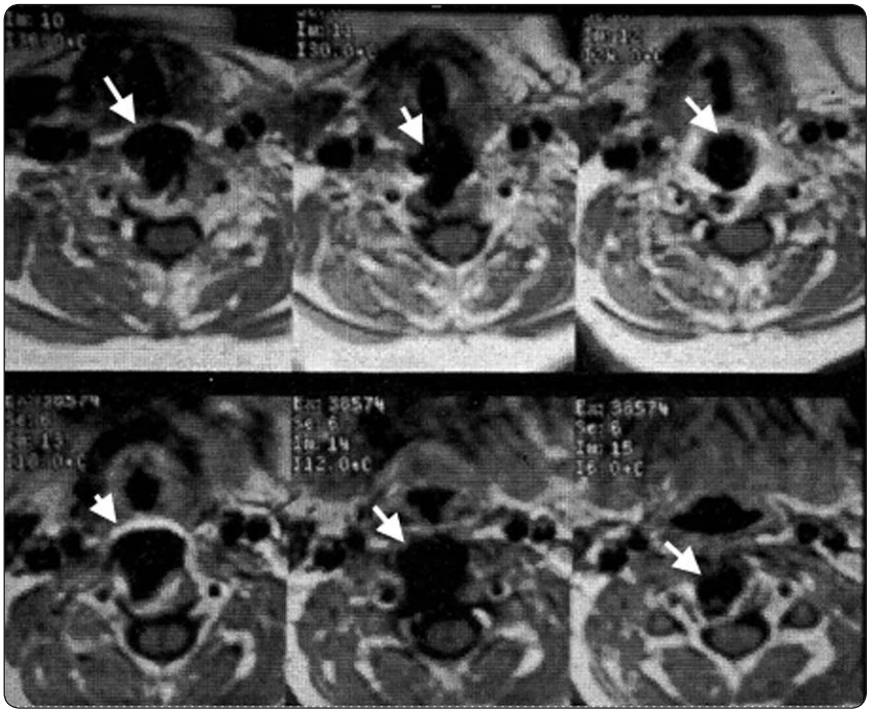


Figure 4. Postoperative magnetic resonance image of the same patient showing complete excision of tumor and the bone cement used to fill the vertebral body defect (Note the arrows referring to bone cement filling the vertebral body defect).

Discussion

Unlike metastasis, primary bone neoplasms of the spine are uncommon representing approximately 5% of all primary bony tumors of the body, and occur in 0.8-8.0 per 100,000 population.[4,15] The cervical spine is the least commonly involved spinal region (approximately 14%) of primary spinal tumors.[15]

In this study, the mean age of patients with benign tumors was 29.3 years while it was 47.5 years in those with malignant tumors. This result is consistent with many reported studies which confirmed the younger age of benign spinal osseous tumors in comparison to the rather older malignant tumors.[3,9]

In Di Lorenzo's series[8] of 38 cases, the mean age of patients with benign and malignant tumors was similar (38 and 54 years, respectively). Likewise, Bohlman et al,[2] reported that the mean age of 23 patients with benign and malignant tumors was 30 and 55 years, respectively.

Primary cervical spine tumors may become symptomatic with various complaints ranging from neck pain, limited neck movement, occipital neuralgia, brachialgia, hand parethesia and mono or quadriparesis.[2,4] These complaints should alert physicians to proceed with radiological evaluation to delineate bony blastic or lytic lesions and the extent of local invasion and to determine if there

is metastasis or spinal instability.[4,9] Patients should be evaluated with plain radiography, CT, MRI, and bone scans.[2,4,9] In this series, one patient with hemangioma presented with neck pain only, 3 patients presented with pain and radiculopathy, and 5 patients presented with pain and myelopathy. In the current study of 8 primary osseous subaxial cervical tumors, the most common benign tumor was aneurysmal bone cyst (2/4=50%). The other 2 benign tumors were hemangioma and osteoblastoma. No predominant malignant tumor was noted. The 4 malignant tumors were giant cell tumor, plasmacytoma, chordoma, chondrosarcoma. In the reported series of 18 cervical osseous tumors, Kelley et al,[15] reported that aneurysmal bone cysts were the most common primary tumor (3/5=60%) while chordomas were the most common malignant tumor (6/13=42.2%) followed by plasmacytoma/ multiple myeloma (3/13=23%).

All of the 8 patients were managed surgically, the aim of which was complete excision of the tumor and stabilization of spine if needed. Consequently, various types of fusion procedures were performed either via anterior, posterior or combined approaches in 7 patients. Di Lorenzo et al,[8] managed nine cases with radical resection and 29 cases with conservative resection. Surgical stabilization was achieved with bone grafts and halo braces in 12 cases. All patients with malignant tumors underwent postoperative radiotherapy. Bohlman et al,[2] managed four patients with biopsy and radiation and 18 patients with excision. Nine patients underwent postoperative radiation.

All the 4 patients with benign tumors are neurologically intact and doing well (Frankel E). One of the two patients with aneurysmal bone cysts who underwent posterior laminectomy developed a recurrence and was reoperated. No recurrence was apparent at late follow-up. In contrast, 2 out of 4 patients with malignant cervical osseous tumors had a residual tumor due to infiltration of vital neuro-vascular structures and died within (12-16 months). The 3rd patient with plasmacytoma died 2years postoperatively from progression to multiple myloma despite local tumor control. On the other hand, the patient with giant cell tumor was reoperated for excision of the local recurrence because she had no evident metastasis with good general condition and she is neurologically intact without recurrence at last follow up 3years. Thus the mortality rate of malignant tumors was very poor (75%) within 3 y postoperatively despite adjuvant therapy.

In Bohlman's series[2] (23 cases), eight of 10 patients with primary malignant tumors died after a mean follow-up of 5 years. Only one patient with a benign tumor died. Twelve patients were pain free and developed solid fusion. Di Lorenzo et al,[8] (38cases) reported a mortality rate of 50% with a 5-year follow-up and of 77% with a 10-year follow-up. All patients with benign tumors had good neurological outcomes and were stable except for one case.

For primary malignant tumors of the spine adjuvant chemotherapy and/or radiotherapy is recommended. The choice of these two modalities is determined by several factors including risk of local relapse and metastasis, resection margins, and the tumor histology, which may indicate the relative chemo- and radiosensitivity.[20]

Intralesional piecemeal excision of malignant osseous spinal tumors is associated with high recurrence rates.[2,3,8,20] Consequently, margin-free en bloc resection[3] and ultimately total en bloc spondylectomy has been described to achieve complete tumor resection and oncologic cure of primary malignant vertebral tumors of the thoracolumbar spine.[19] Regarding cervical spine, only marginal or intralesional piecemeal resection was feasible because of the proximity of the tumor to vital anatomic structures, such as neural elements and the vertebral arteries. However, few case reports has been published about total en bloc spondylectomy for cervical spine.[6,18] It is technically demanding procedures with dissection and preservation of the vertebral arteries and nerve roots.[6,18]

Although the surgical management of 7 out of 8 primary osseous cervical tumors in this series was termed total excision, Really it is classified as marginal piecemeal resection with cervical instrumentation and fusion rather than total en bloc resection. This surgical interference is effective for benign tumors. However, for malignant neoplasms the results are poor (75%) mortality within 3y. Total en bloc spondylectomy may offer better results in the future, but it necessitates a longer follow up for establishing its safety and efficacy.

The surgical management of the 8 primary

osseous cervical tumors in this series is classified as marginal (or intralesional) piecemeal resection with cervical instrumentation and fusion. This surgical interference is effective for benign tumors. However, for malignant neoplasms the results are poor (75%) mortality within 3 years. Total en bloc spondylectomy may offer better results in the future, but it necessitates a longer follow up for establishing its safety and efficacy.

Conclusion

Marginal piecemeal resection with cervical instrumentation and fusion is safe and effective option in benign osseous cervical tumors. It results in improvement of neck or radicular pain, reversal of neurological deficits and early stabilization of the spine. However, the results are poor for malignant tumors even with adjuvant therapy.

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الملخص العربي

الأورام العظمية الأولية في العمود الفقري العنقي تحت المحور: دراسة سريرية

الخلفية العلمية: ان الأورام العظمية الأولية في العمود الفقري العنقي تحت المحور نادرة و ذات اعراض و انواع مختلفة.

تصميم الدراسة: دراسه استعاديه سريرية وصفية

الغرض: تقييم نتائج التدخل الجراحي للأورام العظمية الأولية في العمود الفقري العنقي تحت المحور

المرضي والطرق: شملت الدراسة ٨ مرضي يعانون من أورام عظمية أولية في العمود الفقري العنقي تحت المحور في مستشفي المنيل الجامعي بالقصر العيني خلال الفترة بين عامي ٢٠٠٧ و ٢٠١٢, و تم متابعة الحالات لمدة متوسطها ٢٤,٤ شهر و تم تقييم النتائج بواسطه معامل فرانكل للحاله العصبية و المقياس المرئي التناظري للالم.

النتائج: كان عدد المرضي متساوي من الجنسين , و كان متوسط اعمارهم ٣٨,٤ سنوات. و تبين ان نصف الاورام كان حميدا و النصف الاخر خبيثا. كانت الام العنق موجوده لدي جميع المرضي و عاني معظمهم من اعتلال النخاع ثم اعتلال جذور الأعصاب. جري استئصال جسم الفقره المصابه اماميا مع التثبيت في ٤ حالات, و تم التدخل الجراحي خلفيا في حالتين, و تم التدخل الجراحي اماميا و خلفيا معا في الحالتين الاخريين.

و بعد الجراحة تحسن تقدير معامل فرانكل في ربع الحالات بينما استقر في باقي الحالات مثل قبل الجراحه, و تحسن متوسط المقياس المرئي التناظري من ٥ قبل الجراحة الي ٢,٤ بعد الجراحة. و تحسنت حالة المرضي الأربعة المصابين بأورام حميده بينما توفي ٣ مرضي من الأربعة المصابين بأورام خبيثة خلال ٣ سنوات.

الاستنتاج: ان الاستاصال المجزأ مع تثبيت الفقرات علاج امن و فعال للأورام العظميه الأولية في العمود الفقري العنقي تحت المحورولكن نتائجه سيئة بالنسبه للأورام الخبيثه.