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Case Report

# Giant Cell Reparative Granuloma of the Axis

## Aksis'in Dev Hücreli Reperatif Granüloması

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**ABSTRACT**

Giant cell reparative granuloma (GCRG) is a rare, benign fibroosseous lession. It typically arises in the mandible and maxilla, and less frequently in the skull bones. We report a case of GCRG of the axis, which is the first to be reported in the literature. A 35-year-old man was admitted to our clinic with the complaint of pain at his neck. There was no neurological deficit. CT and MRI showed a lesion destructing the body of the axis. Biopsy specimens were taken through the transoral-transpharyngeal route. Histopathological diagnosis was GCRG. The lesion was removed subtotally by the same route. We filled the tumor cavity with a bone graft and the patient was discharged with a halo brace without any neurological deficits. The follow-up CT revealed one year after the surgery showed sclerosis at the tumor site. The etiopathogenesis of GCRG is still controversial and the differential diagnosis, especially from giant cell tumor of bone is quite difficult. The treatment of choice for these lesions is complete surgical removal. Some authors recommend radiotherapy if total removal fails.

**KEYWORDS:** Giant cell reperative granuloma, Axis, Recurrence

**ÖZ**

Dev hücreli reparatif granüloma (DHRG) nadir görülen iyi huylu fibroosseoz bir lezyondur. Tipik olarak mandibula ve maksillada daha az sıklıkta ise kafa kemiklerinde görülür. Biz literatürde ilk defa aksisin DHRG sini bildiriyoruz. 35 yaşında erkek hasta kliniğimize boyun ağrısı şikayeti ile geldi. Nörolojik defisiti yoktu. CT ve MR da axis cimini destrükte eden lezyon saptandı. Transoral-transfaringeal yol ile alınan biopsi sonucu DHRG olarak saptandı. Lezyon aynı yaklaşım kullanılarak subtotal eksize edildi. Tümör kavitesi kemik greft ile dolduruldu ve hasta nörolojik defisiti olmadan halo ortez ile taburcu edildi. 1 yıl sonra çekilen kontrol CT de tümör bölgesinde skleroz gözlendi. DHRG nin etyopatogenezi hala tartışmalıdır ve özellikle kemiğin dev hücreli tümöründen ayırıcı tanısı oldukça zordur. Tedavi yaklaşımı lezyonun total cerrahi eksizyonudur. Bazı otörler total eksizyonun yapılamadığı durumlarda radyoterapi önermektedir

**ANAHTAR SÖZCÜKLER:** Dev hücreli reparatif granüloma, Aksis, Rekürrens

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## INTRODUCTION

Giant cell reparative granuloma (GCRG) of the axis is a rarely seen nonneoplastic fibro-osseous lesion (2,6,7,8,11). It typically involves the mandibula and maxilla but it has recently been reported that it can also be seen on the bones of the cranium and base of cranium (3,4,6,7,12,13). GCRG is difficult to differentiate from giant cell tumor (GCT); however, differential diagnosis is possible with the developing histopathological criteria. There a few reports suggesting it can also be seen on the spine but there are no reports regarding its localization on the axis. (9,14,15,17).

The aim of the treatment is total excision of the lesion. Many authors suggest radiotherapy for cases where the lesion cannot be totally excised. The recurrence rates vary between 10% and 75% (1,4,7,8,10,18).

In this case report, we are presenting a case with GCRG localized on the axis. The lesion was only subtotally excised because of the localization and the patient could not receive radiotherapy as he had fusion material. Despite these factors, no recurrence occurred after a year of follow up.

## CASE REPORT

A 35-year-old male patient applied to our clinic with neck pain, which increased with cervical movements. The patient's history revealed that he had fallen down from a high place and landed on his neck 3 months ago. However, he had no neurological deficits. Laboratory test results for calcium, phosphorus, and alkaline phosphatase were within normal ranges. A radiolucent complexion and irregular axis contour were seen on lateral cervical x-ray.

A hypodense lytic lesion without sclerosis involving the axis body and odontoid was seen on the upper cervical Computerized Tomography. (Figure 1A,B) Cervical Magnetic Resonance imaging showed that the lesion caused destruction of the axis and odontoid, but it did not cause compression of the spinal canal. (Figure 2A,2B)

Biopsy material was obtained via the Transoral-transpharyngeal approach, and the pathological evaluation revealed that the lesion was a GCRG. (Figure 3) During the reoperation of the patient, a well-vascularized, gray-red colored, soft tumor tissue that invaded odontoid and body of axis was seen when the mucosa and muscular layer were

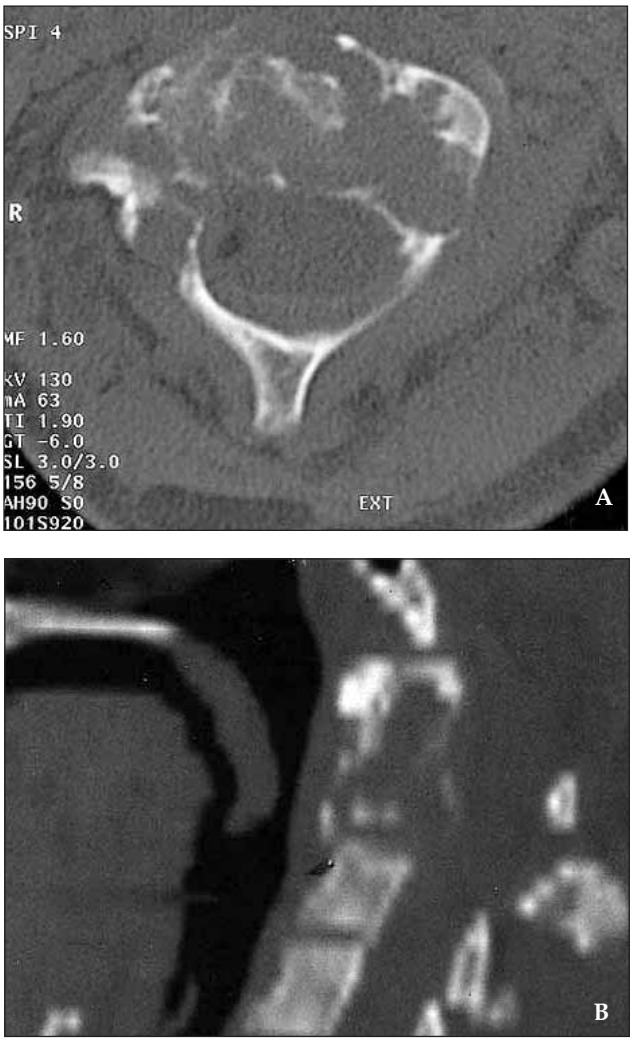
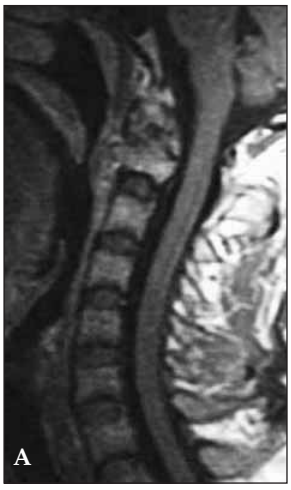



***Figure 1:*** ***A*** *(transverse),* ***B*** *(sagittal) CT without contrast revealing hypodense-lytic lesion involving the body, lateral masses and the pedicle of the axis.*

splitted with the transpharyngeal approach. The tumor was excised subtotally. The tumor cavity was filled with demineralised bone matrix flex. The cervical spine of the patient was immobilized with a halo brace. The patient was discharged from the hospital on the 6th postoperative day without any neurological deficits.

Histopathological examination of the tumor confirmed the diagnosis of GCRG. The halo brace was removed 3 months after the operation. CT (Computerized Tomography) and MR (Magnetic Resonance) imaging performed 1 year after the operation revealed that previous tumor localization was sclerosed and there were no recurrences in the tumor cavity. (Figure 4A,B)



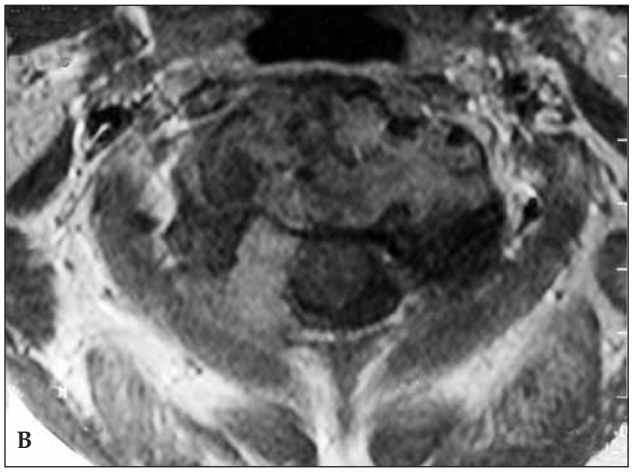




***Figure 2:*** ***A*** *(sagittal) T1-weighted MRI. Without contrast image revealing hypo and hyperintense lesion on the body of the axis. The protrusion into the pharynx and obliteration of the anterior subarachnoid space are also seen.* ***B*** *(axial) Postcontrast axial imaging showed the lesion with heterogenous intensity.*

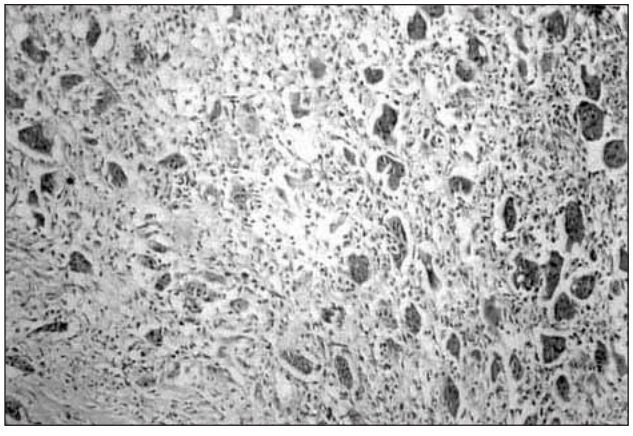
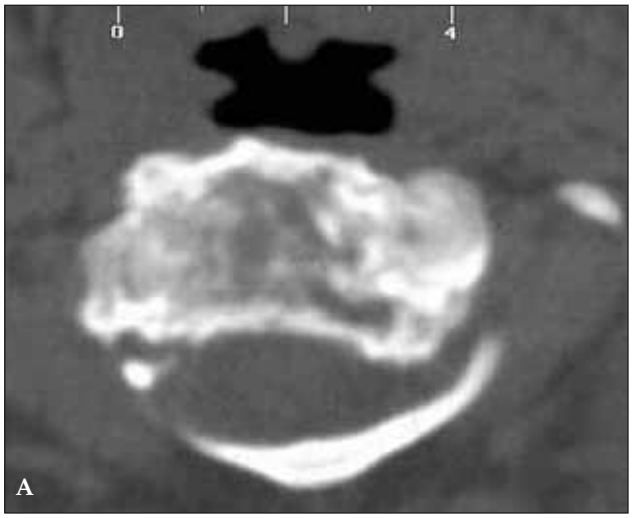


***Figure 3:*** *Photomicrograph showing multinucleated giant cells in the fibrous stroma. The nuclei of the giant cells are few and disorderly. The benign character of the stroma is also seen. H.E. 160X.*



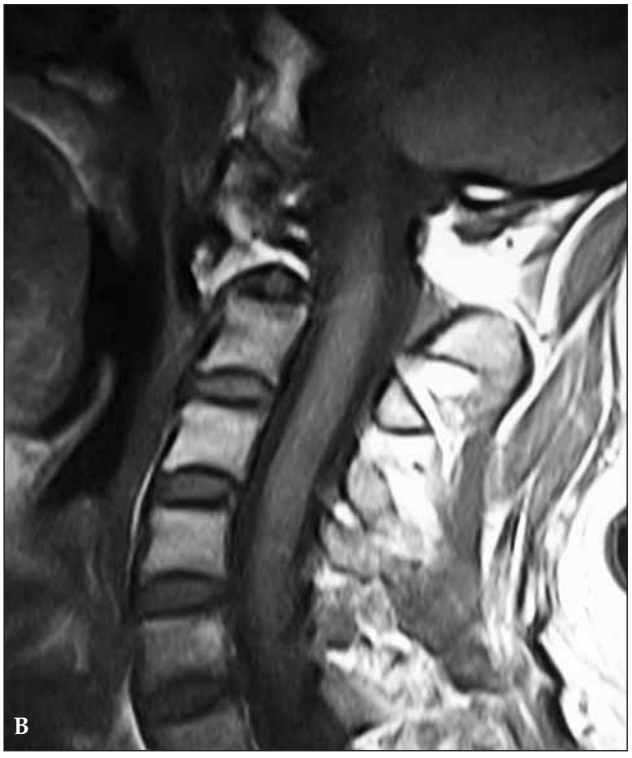




***Figure 4:*** ***A*** *(axial) CT 1 year after operation revealing sclerosis at the tumor site.* ***B*** *(sagittal) Postcontrast sagittal MRI showed no recurrence at the tumor site and no compression of the anterior subarachnoid space .*

## DISCUSSION

The GCRG definition was first used by Jaffe in 1953 for a benign fibro-osseous lesion of maxilla and mandibula (11). Before this definition, lesions with similar histopathological features were defined as GCT. Although the most important histopathological feature common to both lesions is multinuclear giant cells, the giant cells of GCRG are more cumulated,

have less nucleus number and more cytoplasmic area compared to GCT (2,7). New bone formation, hemorrhage, and hemosiderin are other differentiating histopathological characteristics of GCRG. After the development of these diagnostic criteria, Katz and Hirsl showed the temporal bone localization of GCRG in 1974, which was the first report indicating that these lesions could be localized in bones other than jawbones (7). The same authors reevaluated the nine patients who were diagnosed as temporal bone GCRG clinically and histopathologically and determined that only six of these patients' pathological diagnoses were GCRG. Afterwards, the number of case reports about GCRG localized in the orbita, sphenoid bones, paranasal sinuses, cranial base bones, and rarely the spine increased. There are few reports describing GCRG localized in the cervical vertebra so far and the case reported here is the first one localized in the axis (14,17).

While most authors suggest that an intra-osseous hemorrhage or periosteum reaction following trauma is responsible for GCRG development (8,11,12,13), others claim that developmental abnormalities and inflammation are the causes underlying GCRG (10). Although trauma is the most blamed factor, most of the cases in the literature do not have a trauma history. In patients with a history of trauma, the duration between the trauma and diagnosis varies from a few months to years. Our case had a history of severe cervical trauma and the period between the trauma and the diagnosis was only 3 months. The fact that diagnosis of GCRG was made shortly after the trauma and cervical CT image showed that the cortical bone was relatively protected in this case supports the idea that GCRG can develop following trauma.

Other osteolytic lesions such as GCT, aneurismal bone cysts, Brown tumor related with hyperparathyroidism, chondroblastoma, fibrous dysplasia, and osteosarcoma must be kept in mind during the differential diagnosis of GCRG. (1,2,5,7,16) Differential diagnosis of GCRG, which is a non-neoplastic benign lesion, from GCT, which has higher recurrence and metastasis rates, is very important. Lesions other than GCT and Brown tumor can be easily differentiated from GCRG both histopathologically and radiologically. Serum calcium, phosphorus, and alkaline phosphatase levels help the differential diagnosis of Brown tumor. When GCRG radiological imaging studies were evaluated, there were different reports regarding the sequence properties of T1, T2 with MR imaging and contrast absorbance. The reason for this difference may be related to the age of intra-osseous hemorrhage. As there are controversial opinions on MR images, a careful pathological evaluation is the most important factor especially for differential diagnosis of GCRG from GCT (15,18).

The treatment of GCRG is total resection of the lesion if possible. Most authors suggest low-dose radiotherapy if total excision cannot be performed (4,13,15,18). The recurrence rates for GCRG are 10-15% in most studies but there are studies showing recurrence rates as high as 69-75% (1,4,7,8,10,18). In this case, subtotal excision was performed as the lesion was in proximity to the neurovascular structures, and radiotherapy was not administered because of the fusion material that was embedded for stabilization. Despite these factors, there was no recurrence 1 year after the operation.

## CONCLUSION

Although GCRG is traditionally seen in the mandibula and maxilla, we want to emphasize that it can be seen in the axis. The treatment of choice for these lesions is complete surgical removal and some authors recommend radiotherapy if total removal fails. Complete removal may not be accomplished in some locations and radiotherapy cannot be administered because of the fusion material as in our case.

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