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Case Report
Novel techniques for solitary atlas osteochondroma: a case report and literature review

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Abstract: Osteochondroma is a common benign bone tumor that is rarely seen in the spine, especially in the atlas. Although most solitary atlas osteochondromas have no symptoms, some exostosis may cause severe clinical symptoms that need treatment within the spine. Here, we report a 21-year-old male who presented with apsychia as well as numbness in his right upper and lower limbs for 2 months. The patient reported a history of neck trauma 10 years ago. He received a posterior laminectomy without reconstruction later, and the symptoms improved immediately. During a 32 month follow-up, there was no recurrence of the osteochondroma. Novel techniques for the treatment of this case were applied: simulated surgical resection using 3-Matic 9.0 software, 3D printed model, 3D Digital Image Microscopy, and piezoelectric surgery. These novel techniques provided significant benefits to the patients, the surgeon, and medical education.

Keywords: Osteochondroma, atlas, 3D printing, 3D microscope

Introduction

Osteochondroma, also known as exostosis, is a common benign bone tumor comprising cortical and medullary bone with a hyaline cartilage cap, accounting for 20-50% of all bone tumors [1]. These tumors consist of two types: solitary osteochondroma and multiple exostosis [2, 3]. It is commonly found in the extremities but rarely seen in the spine [4]. The occurrence of spinal osteochondromas is more frequent in multiple exostosis. Moreover, the axis is most often involved in cervical vertebra solitary osteochondromas, and is less frequent in the atlas. Although most solitary atlas osteochondromas have no symptoms, some exostosis may cause severe clinical symptoms that need treatment [5]. The literature reports only distinguished cases of solitary atlas osteochondroma; however, the characteristics are still unknown. Herein, we report a solitary atlas osteochondroma causing myelopathy and the novel techniques used for its treatment. Then we review the current literature reports of solitary atlas osteochondromas and analyze the demographic characteristics, clinical presentation, and the treatment strategies.

Case report

We report a 21-year-old male who presented with apsychia as well as numbness in his right upper and lower limbs for 2 months. Two months ago, the patient had a sudden syncope while walking and recovered consciousness after approximately 3 minutes, and subsequently felt numbness in the right limb. The patient reported that he had a history of neck trauma 10 years ago and has experienced headaches and dizziness after the trauma.

Upon examination, a forced position was found, and the neck could not turn left. The superficial sensation of the right limb decreased. The muscle strength of the limbs was normal. Knee tendon and Achilles's tendon reflexes were hyperactive. Pathological signs were negative. The JOA was 8.

Conventional anterior-posterior and lateral X-ray of the cervical spine provided limited information (**Figure 1**). However, computed tomography scans showed an extradural osseous protrusion projecting into the spinal canal in continuity with the posterior arch of the atlas.

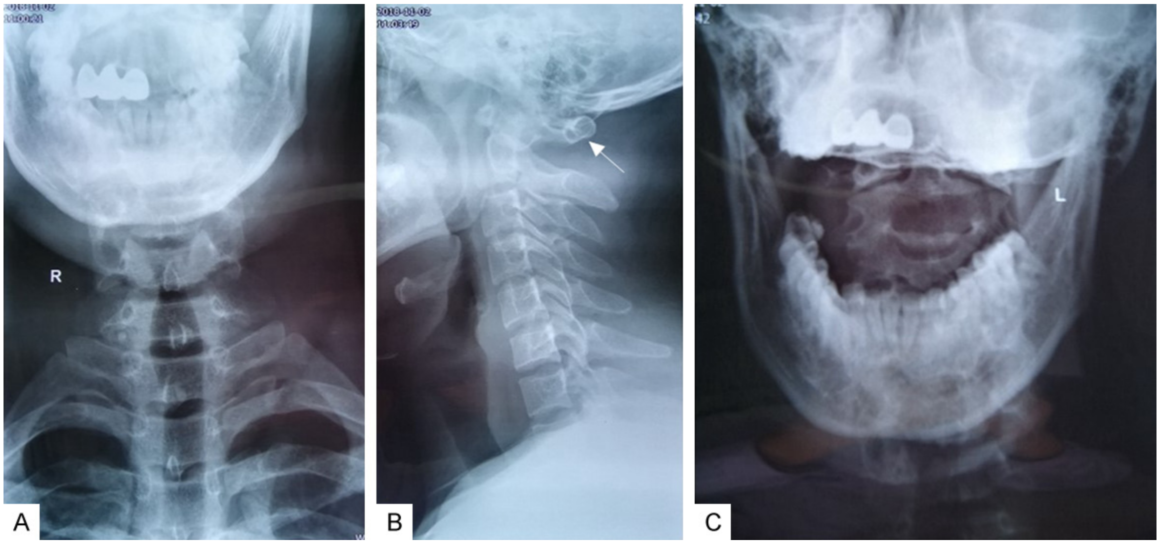

Figure 1. The pre-operation X-ray. Conventional X-ray of the cervical spine provided limited information. A. Anterior-posterior view; B. Lateral view; C. Atlantoaxial vertebral opened-mouth position.

Magnetic resonance imaging of the spine showed a large extradural mass lesion arising from the atlas posterior arch leading to spinal cord compression (**Figure 2**). Electrophysiological testing indicated normal nerve conduction velocity and evoked potential amplitudes within his upper extremities.

The patient was given a clinical diagnosis as having "solitary atlas osteochondroma" because no other osteochondroma was found.

For a more accurate osteotomy, we used the Mimics 17.0 software (Materialize, Belgium) for the 3D reconstruction based on preoperative CT scans (**Figure 3**). The atlas and the osteochondroma were reconstructed and exported as a ".stl" file. The ".stl" files were processed using the 3-Matic 9.0 software (Materialize, Belgium) for surgery simulation (Supplementary Material). Then the ".stl" file was processed by the 3D printer to fabricate the actual 3D model in acrylate resin using stereolithography. Then, the 3D model was thoroughly washed and vacuum dried. After sterilization with ethylene oxide, the 3D printed templates were stocked in plastic bags. The patient underwent a posterior laminectomy without reconstruction and radical resection of the tumor using piezoelectric surgery under a 3D Digital Image Microscope (**Figure 4**). In order to prevent nerve injury during the process of turning over, we used nasal intubation under awakening and drug administration after turning over. Intraoperative electrophysiological monitoring showed no significant abnormalities.

The postoperative course was uneventful, and the symptoms improved immediately after surgery. Histopathological analysis confirmed the diagnosis of osteochondroma (**Figure 5**). At a 3-month follow-up, the patient had mild numbness of the right upper limb, and the JOA was 16. The lesion was found to have been completely removed during magnetic resonance imaging, and spinal cord compression was relieved (**Figure 6**). During a 32-month follow-up, there was no recurrence of the osteochondroma.

Discussion

A variety of signs and symptoms could be caused by osteochondromas within the spine, accounting for approximately 1% to 4% of all the clinically diagnosed osteochondromas. The most common occurrence of osteochondromas is cervical [6]. However, osteochondroma in the atlas are rarely reported, especially the solitary osteochondroma. Paine et al. first reported the solitary atlas osteochondroma in 1956 and resected the tumor via a laminectomy without reconstruction [7].

We performed a thorough review of the English medical literature using PubMed, Web of science, and Ovid MEDLINE and found 30 report-

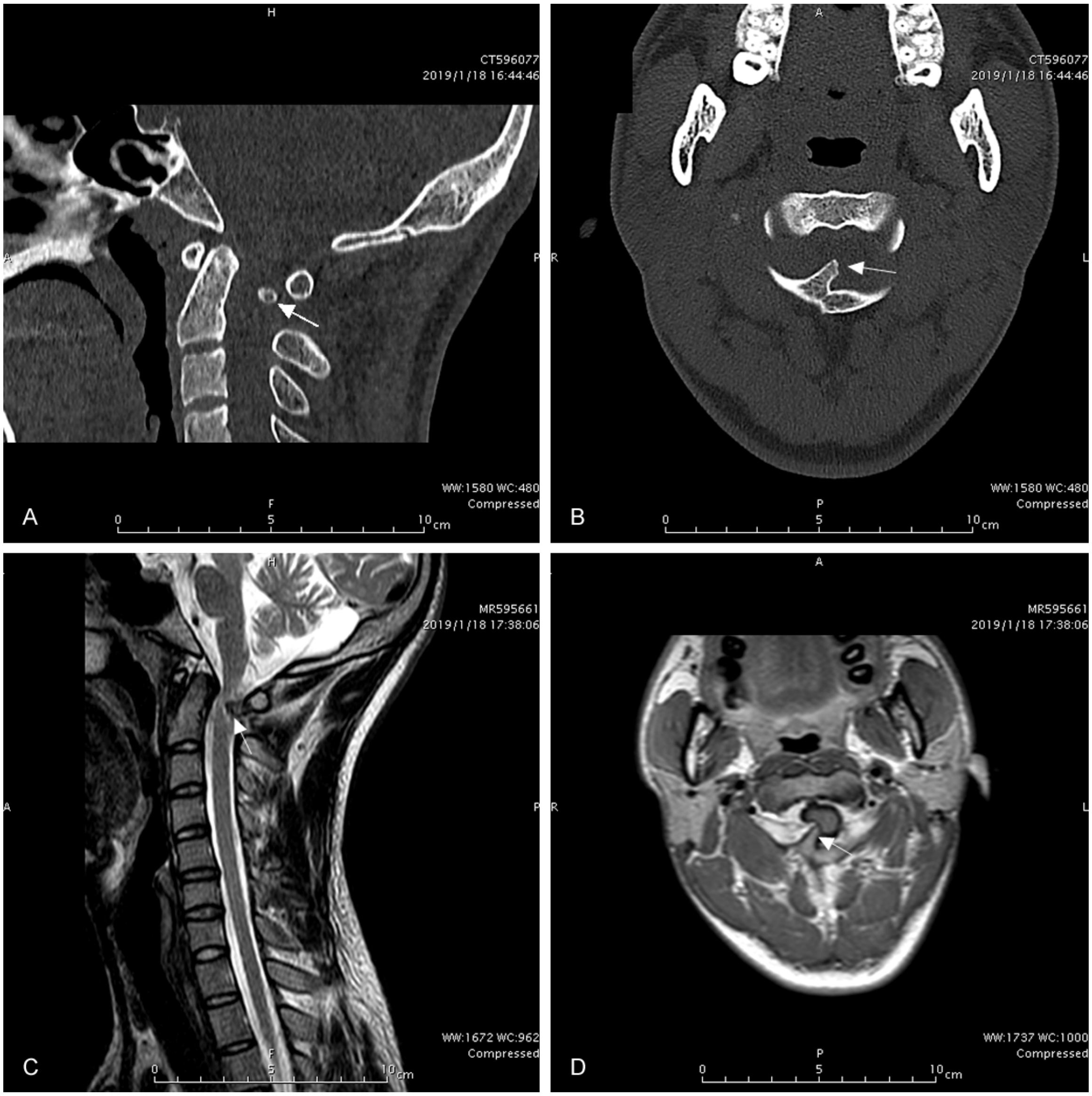

Figure 2. CT and MRI scan of the atlantoaxial. The CT and MRI scan shows an extradural osseous protrusion projecting into the spinal canal in continuity with the posterior arch of the atlas leading to spinal cord compression (The white arrows show the location of the tumor). A and C. Sagittal view of the atlantoaxial vertebral; B and D. Cross-sectional view of the atlas.

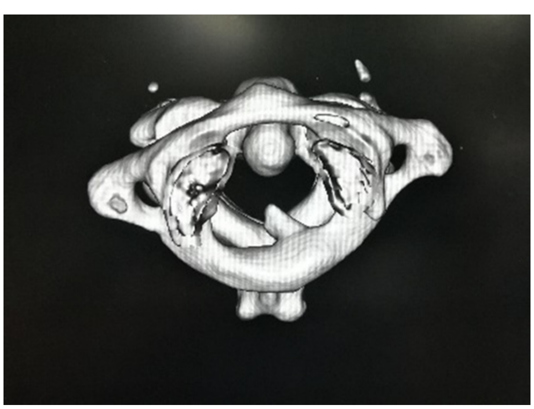

Figure 3. The 3D reconstruction based on preoperative CT scans. 3D rendering visualizing the location of the extradural osseous.

ed cases of solitary atlas osteochondroma in addition to our case (**Table 1**) [7-30]. The demographic pattern revealed the onset of these lesions at a mean age of 39.60±18.38 yrs with a sex demographic of 16 males to 14 females. The primary clinical manifestations were myelopathy; however, some cases were different due to the location of the lesions and sleep

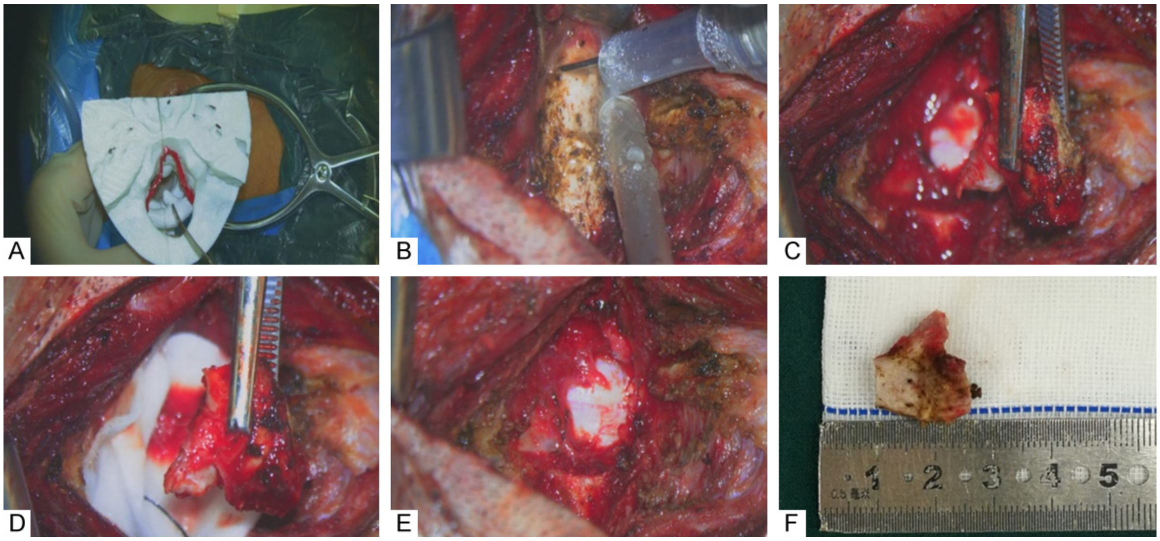

Figure 4. The surgical resection process of the osteochondroma. The osteochondroma was completely resected with the help of sterilized 3D-printed model and piezosurgery. A. Sterilized 3D-printed model showed the exact location of the osteochondroma, B. The resection of the posterior arch of the atlas with piezosurgery; C and D. Complete osteochondroma resection; E. After tumor resection the spinal cord was no longer compressed and the dura mater was intact; F. Image of the resected osteochondroma.

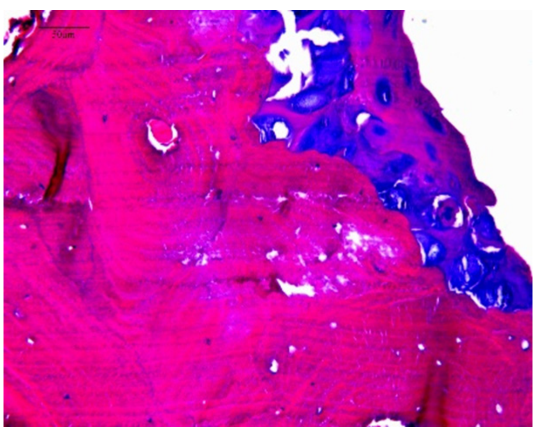

Figure 5. Histopathological analysis confirmed the diagnosis of osteochondroma. Hematoxylin and Eosin staining of the tumor (original magnification 100×) showed the cartilaginous cap and underlying bone that confirmed the osteochondroma diagnosis.

apnea. The most common lesion distribution location was in the posterior arch. In addition to our case, there were three cases with a history of trauma, which may confound the diagnosis. With a 27.26±29.17 month follow-up, there was no tumor recurrence reported. Moreover, resection of the posterior arch of the atlas does not affect stability; therefore, the most common treatment of a solitary atlas osteochondroma is a laminectomy without reconstruction. Depending on the tumor's location, different surgical procedures such as a lateral mass and anterior excisions are utilized. In our case, the lesion started from the atlas posterior arch. Radical resection of the tumor was accomplished using a posterior laminectomy without reconstruction.

Different from previous cases, some novel techniques for the treatment of this case were applied: simulated surgical resection using 3-Matic 9.0 software, a 3D printed model, 3D Digital Image Microscopy, and piezoelectric surgery. These novel techniques had benefits to the patient, the surgeon, and medical education.

Simulated surgical resection using 3-Matic 9.0 software

We imported preoperative CT data into the Mimics software, generating 3D images of the atlantoaxial vertebra and tumors. Then we measured the distance between the base of the osteochondroma and the midline of the atlas posterior arch, which was 22.05 mm from midline to the left and 10.03 mm from midline to the right. Using this information, we determined the extent of the excision. Then the files were processed using the surgery simulation 3-Matic 9.0 software. The data mentioned above was shown to the patient and his family,

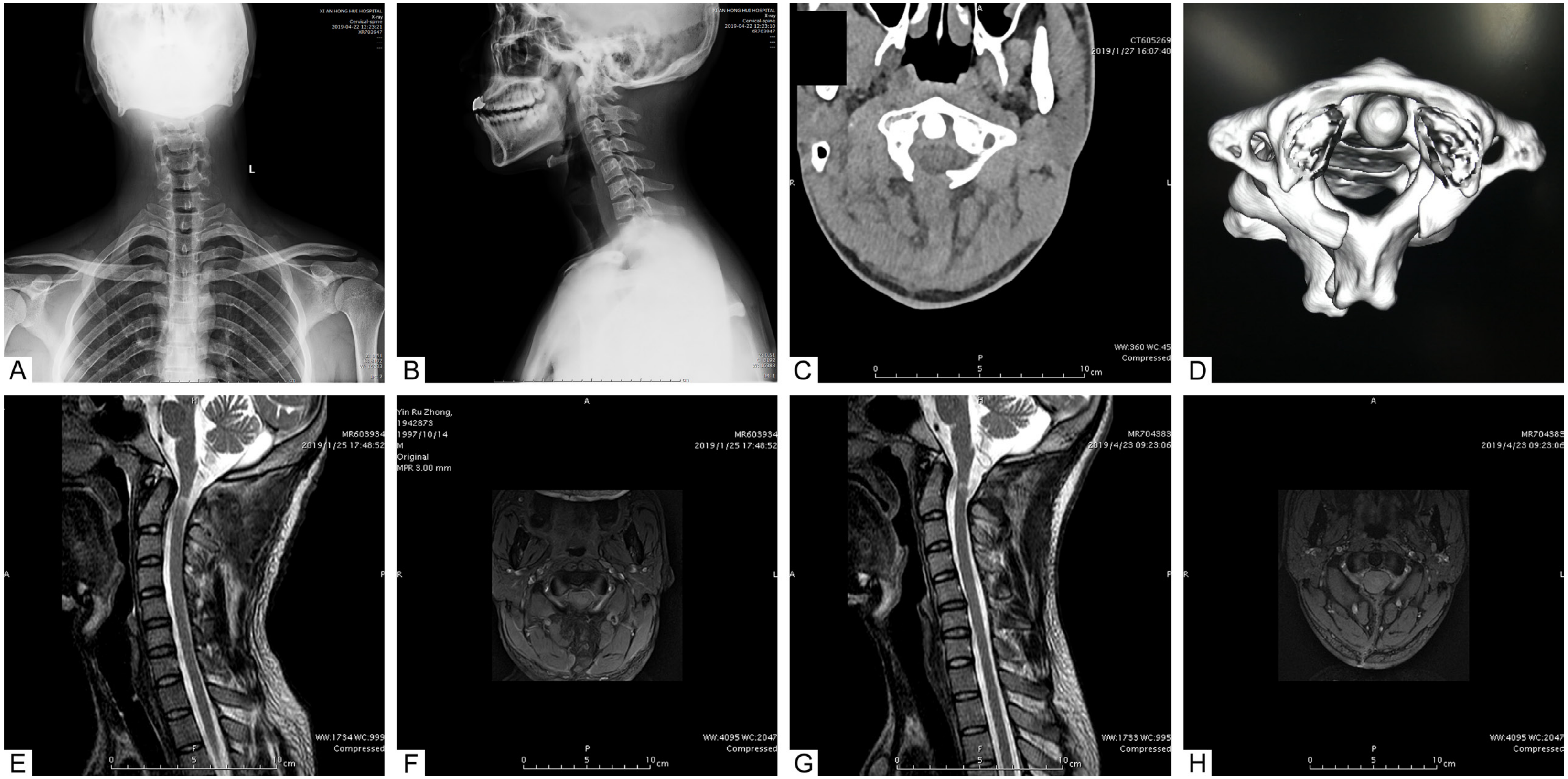

Figure 6. Post operative imaging examination showed complete resection of the osteochondroma. The lesion was confirmed to be completely removed via magnetic resonance imaging as well as compression of the spinal cord was relieved. A and B. X-ray showed no significant change compared with the preoperative images; C and D. CT scan and reconstruction 3 days post operation revealed complete resection of the osteochondroma and the resection range of posterior arch of the atlas; E and F. MRI images 3 days post operation revealed that the spinal cord compression was relieved after the tumor resection. G and H. 3 months post operation imaging shows significant recovery of the spinal cord and surgical incision after tumor resection.

Table 1. Summary of previously reported cases and present case

Author	Journal	Year	Age	Sex	Clinical manifestation	Symptom duration	History of trauma	Location of the lesion	Resection	Recon-struction	Clinical Outcomes	Follow-up	Recur-rence
Paine [7]	Proceedings of the Royal Society of Medicine	1956	20	F	Weakness in right limbs	17 months	19 months ago	Posterior arch	Laminectomy	No	Near complete resolution	N/A	N/A
Mitsumori [8]	No Shinkei Geka. Neurological Surgery	1975	54	F	Neck pain	1.5 years	No	Atlanto-odon-toid joint	Anterior excision	No	Complete resolution	N/A	N/A
Wu [9]	Clinical Orthopaedics and Related Research	1978	54	F	Pain in neck and right should	7 months	No	Lateral mass	Anterior excision	No	Complete resolution	6 years	No
Julien [10]	Journal of Neurology, Neurosurgery, and Psychiatry	1978	28	F	Brown-Sequard syn-drome	4 years	No	Posterior arch	Laminectomy	No	N/A	N/A	N/A
Lanzieri [11]	Journal of Computer Assisted Tomography	1985	19	M	Weakness in all extrem-ity	N/A	No	Posterior arch	Laminectomy	No	N/A	N/A	N/A
Slavotinek [12]	Neuroradiology	1991	57	F	Dysphagia, left 10 and 12 cranial nerve palsies	2 years	No	Lateral mass	Lateral excision	No	Near complete resolution	1 years	No
Calhoun [13]	Surgical Neurology	1992	43	M	Weakness in left limbs and hyper reflexia	1 year	No	Posterior arch	Laminectomy	No	Near complete resolution	6 week	No
Lopez-Barea [14]	Clinical Orthopaedics and Related Research	1994	31	M	Neck pain and hyper reflexia	N/A	No	Posterior arch	Laminectomy	No	N/A	N/A	N/A
Morikawa [15]	Clin Imaging	1995	21	M	Numbness of both hands and hyper reflexia	6 months	6 months ago	Posterior arch	Laminectomy	No	N/A	N/A	N/A
Khosla [16]	Spine	1999	39	M	Weakness in upper extremity	N/A	No	Left C1 pedicle	Hemilaminectomy	No	Complete resolution	35 months	No
Khosla [16]	Spine	1999	45	M	Weakness and hyperto-nia in left limbs	2 years	No	Posterior arch	Hemilaminectomy	No	Complete resolution	33 months	No
Khosla [16]	Spine	1999	39	M	Weakness and hyperto-nia in bilateral limbs	N/A	No	Lateral mass	Hemilaminectomy	No	Complete resolution	35 months	No
Sharma [17]	Journal of Neurosurgi-cal Sciences	2002	40	M	Quadraparesis	1 year	No	Posterior arch	N/A	N/A	N/A	N/A	N/A
Kroppenstedt [18]	Surgical Neurology	2002	61	M	Quadraparesis	3 years	No	Posterior arch	Laminectomy	No	Near complete resolution	1 year	N/A
Yoshida [19]	Acta Otolaryngol	2006	61	F	Sleep apnea	8 years	No	Anterior arch	Anterior excision	No	Complete resolution	6 months	No
Ozturk [20]	Acta Orthopaedica Belgica	2007	46	M	Pain and numbness of neck and left arm	1 month	No	Posterior arch	Hemilaminectomy	Yes; C1-2	Complete resolution	6 months	No
Wang [21]	Journal of Clinical Neuroscience	2009	18	F	Dysphagia and sleep apnea	2 years	No	Anterior arch	Anterior excision	No	Complete resolution	10 months	No
Yagi [22]	Journal of Neurosur-gery: Spine	2009	77	F	Neck pain and gait disturbance	5 years	No	C1/C2 articulation	Hemilaminectomy	No	Complete resolution	2 years	No
Schomacher [23]	Acta Neurochirurgica	2009	62	M	Neck and shoulder pain	1.5 years	No	C1/C2 articulation	Hemilaminectomy	No	Complete resolution	10 days	No
Miyakoshi [24]	Neurologia Medico-Chirurgica	2010	58	M	Numbness of both hands and hyper reflexia	10 years	No	Posterior arch	Laminectomy	No	Complete resolution	3 years	No

Er [25]	Asian Spine Journal	2012	42	F	Weakness, pain, numbness and spastic quadriparesis	4 months	10 year ago	Posterior arch	Laminectomy	No	Complete resolution	2 years	No
Rahman [26]	BMJ:Case Reports	2012	16	M	Headache and quadriparaesis	6 months	No	Posterior arch	Laminectomy	No	Near complete resolution	1 years	No
Zaijun [27]	Journal of Spinal Disorders and Techniques	2013	60	F	Pain, dysphagia and hypesthesia	N/A	No	Lateral mass	Anterior excision	Yes; C1-2	Worse	49 months	No
Zaijun [27]	Journal of Spinal Disorders and Techniques	2013	17	F	Pain, dysphagia and hypesthesia	N/A	No	Transverse process	Complete excision	No	Complete resolution	121 months	No
Zaijun [27]	Journal of Spinal Disorders and Techniques	2013	60	F	Myelopathy	N/A	No	Lateral mass	Complete excision	No	Worsening of symptoms	49months	No
Zaijun [27]	Journal of Spinal Disorders and Techniques	2013	26	M	Pain and quadriplegia	N/A	No	Lateral mass	Anterior excision	Yes; C1-2	Complete resolution	33 months	No
Zhang [28]	Skeletal Radiol	2015	19	F	Vertigo, nausea, vomiting and headache	2 days	No	Lateral mass	Lateral mass excision	No	Complete resolution	6 months	No
Sultan [29]	Pediatric Neurosurgery	2016	8	M	Left-sided weakness and hyper reflexia	10 days	No	Posterior arch	Laminectomy	No	Complete resolution	1 week	No
Lotfinia [30]	Spinal Cord Series and Cases	2017	48	F	Left side paresthesia, weakness	6 months	No	Posterior arch	Laminectomy	No	Complete resolution	5 years	No
Zhang	Present case	###	19	M	Right side paresthesia and hyper reflexia	2 months	10 year ago	Posterior arch	Laminectomy	No	Near complete resolution	32 months	No

N/A, not applicable.

which is conducive to preoperative communication. Meanwhile, the anesthesiologist and the surgical assistant were also informed of this preoperative plan, which contributes to surgical cooperation. Furthermore, this preoperative planning data can be used in clinical teaching.

3D printed model

Based on the CT data of the atlantoaxial vertebrae, the 3D printed model was made using a photosensitive resin. Preoperative visualization of the relationship between the atlas and the tumors was showed to the patient and his family. The 3D printed model was then used during the operation after low-temperature plasma disinfection in order to assist the surgical team in visually confirming the extent of the surgical resection.

3D digital image microscope

During the surgery, surgeons can hold a comfortable upright posture to operate and perceive depth by observing the images on the 3D monitor. The eyepiece design significantly reduces the fatigue caused by constant eyepiece observation as well as bending over the surgical field; thus, improving the surgeons' working environment. All surgical participants share the surgeon's first-person perspectives in real-time, which is helpful for surgical cooperation between the surgeon and the assistants. The video stream can be shared in real-time to help with training or the ability to go back and review the surgery. This is beneficial because it allows for real-time clinical teaching or remote consultation. To date, this is the first solitary atlas osteochondroma resection using a 3D Digital Image Microscope.

Piezoelectric surgery

In this case, the lesion basing the posterior arch of the atlas projected into the spinal canal. We needed a safer device to avoid the risk of a dural tear and spinal cord injuries while using a high-speed drill. Piezoelectric surgery is an ultrasonic device that cuts the bone, but does not damage the soft tissue and the durometer. This type of selectivity within a bone scalpel is ideal for spinal surgeries. We used piezoelectric surgery to cut the atlas's posterior arch and remove the osteochondroma without a dural tear. Furthermore, the width of the osteotomy is accurate to 0.5-0.7 mm; therefore, precise resection can be performed according to preoperative planning.

Conclusion

Although spinal cord compression due to a solitary atlas osteochondroma is rare, especial for the atlas, it can result in severe myelopathy. Resection of the posterior arch of the atlas without reconstruction does not affect stability, and the complete removal of the tumor is necessary to achieve decompression and avoid recurrence. Novel techniques could help the precision of the surgery; thus, avoiding the risks of the traditional methods.

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Disclosure of conflict of interest

None.

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