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GRAND ROUNDS

Aneurysmal bone cyst of C2 treated with novel anterior reconstruction and stabilization

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Abstract
Purpose

Aneurysmal bone cysts (ABC) form 1 % of primary bone tumors. Reported incidence rates are no more than 1.4 to 1,00,000. ABC of spine frequently involves posterior elements and commonly affects the lumbar spine (45 %). We present a case of C2 ABC for the challenges it poses due to the rarity of the lesion, tedious to access location, dilemmas relating to the suitable approach for tumor resection and technically demanding stabilization and reconstruction strategy post resection.
Methods Clinical data analysis was performed to discuss a method of novel anterior column reconstruction following resection of a C2 aneurysmal bone cyst in a 8 year old child with anterior and posterior elements being involved.
Results An 8-year-old girl with an aneurysmal bone cyst of the C2 vertebra underwent staged surgery following preoperative embolisation. First a posterior approach tumor excision with posterior instrumented fusion was performed. Following which, using a modified anterior retropharyngeal approach anterior tumor excision and fibular graft reconstruction between the C1 lateral mass and C2 body was performed. Complete tumor clearance and stable reconstruction was successfully achieved in our patient. Patient showed excellent clinical outcome with radiological fusion.
Conclusions Preoperative embolisation in the treatment of ABC has supplemental advantage by reducing blood loss. Modified anterior retropharyngeal approach allows satisfactory clearance for C1–2 lesion and fibular strut graft between the C1 lateral mass and C2 body can provide a stable graft placement with good chance of fusion. Instability and spinal deformity, whether preexisting or post-excision, should be corrected with reconstruction and stabilization to offer best chance of cure in such cases.

Keywords Aneurysmal bone cyst · Cervical spine tumor · C2 vertebra

Case presentation

An 8-year-old girl presented to our clinic with neck pain and stiffness in the neck that she had endured for 3 months. The child had diffuse fullness over the upper 1/3 of right posterior triangle which was non-pulsatile. Cough impulse was absent. Cervical spine tenderness and paraspinal muscle spasm was present over the C1–C3 region on the right side. Movements of the cervical spine were painful and restricted. Neurological examination was normal.

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Diagnostic imaging

The patient was evaluated by an X-ray, CT scan and MRI of the cervical spine. The CT scan of the cervical spine (Fig. 1) showed a large osteolytic expansile lesion involving the anterior body, right lateral mass and right neural arch of C2 vertebra with scalloped margins and showed presence of a cortical break. There was absence of any periosteal reaction or soft tissue extension.

Magnetic resonance imaging of the cervical spine (Fig. 2) displayed a large, expansile, heterogeneous, multilobulated lesion measuring 4 $\times$ 2 $\times$ 2.5 cm involving the C2 vertebra, mostly on the right side. The lesion involved the right lateral mass, pedicle and anterior body with fluid–fluid level signal within the tumor. The tumor also showed thinned out cortex with a soft tissue component as well. The cysts were heterogeneous in appearance according to both T1- and T2-weighted images. The presence of multilobulated cysts containing fluid–fluid interfaces on T2 was characteristic and highly suggestive of the diagnosis of aneurysmal bone. Preoperative angiography and vertebral artery embolisation was performed with Amplatzer vascular plug, nestor fibered coil and PVA particles to minimize blood loss. Through a right femoral access right subclavian artery injection showed a tumor blush in the region of the right C2 vertebral body with multiple feeding vessels from the V2 segment of the vertebral artery and braches from the deep thyrocervical trunk (Fig. 3). The vertebral artery showed a 20 % stenosis in the region of the tumor blush. The blood flow in the basilar, cerebellar and posterior cerebral artery was normal. Left vertebral injection showed normal retrograde filling of the right vertebral artery. The right vertebral artery was embolised across the abnormal segment using Amplatzer vascular plug and Fibered coil. The residual tumor filling was embolised using PVA particles. The thyrocervical trunk was embolised using the PVA particles. The right vertebral artery (v2 segment) was found to be encased by the tumor with narrowing of vessel caliber; however, the flow appeared to be maintained.

Historical review

Aneurysmal bone cysts (ABC) are benign lesions, which nevertheless can behave aggressively [1–3]. Aneurysmal bone cysts (ABC) form 1 % of primary bone tumors and 15 % of primary tumors of the spine [4–6]. Reported incidence rates are no more than 1.4 to 1,00,000. ABC of spine frequently involves posterior elements and commonly affect the lumbar spine (45 %) followed by cervical and thoracic regions [4]. It commonly affects the pediatric age group and can lead to deformity and instability which needs to be addressed appropriately [4, 7, 8].

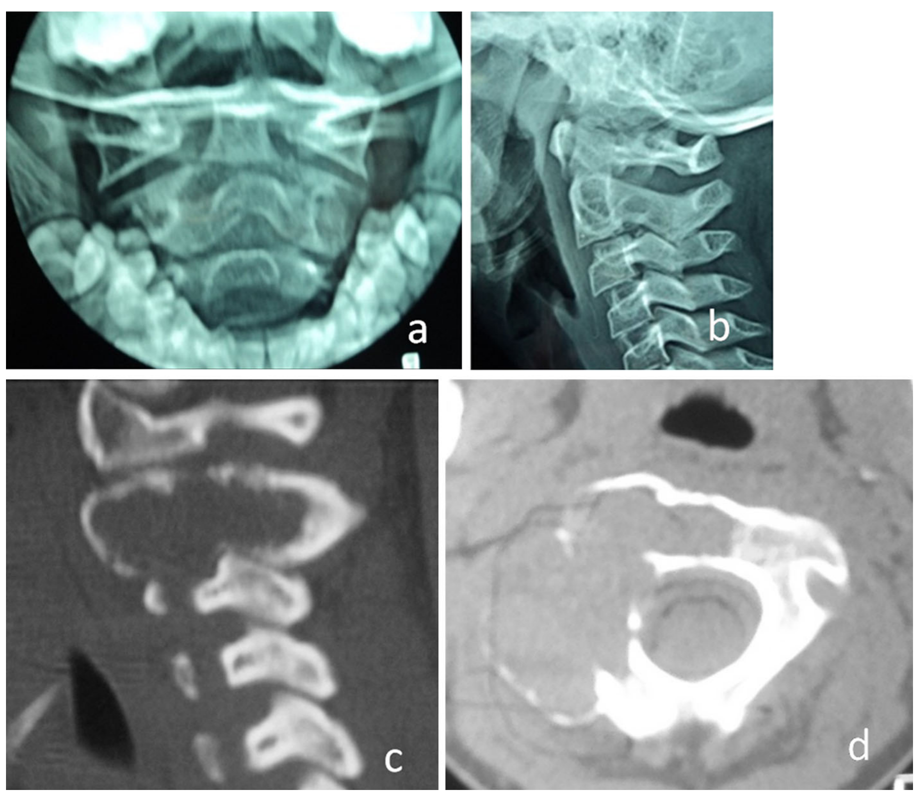

Fig. 1 Pre-operative X-ray and CT images. **a** X-ray cervical spine open mouth view. **b** X-ray cervical spine lateral view, however, no obvious bony destruction is noted on the radiographs. **c** Sagittal section on the CT scan shows an expansile lytic lesion in the C2 vertebra involving the right neural arch, lateral mass and extending into the anterior vertebral body. **d** Axial section through the C2 vertebra shows expansile lytic lesion involving both anterior and posterior elements with thinned out and breeched cortex. The anterior vertebral body involvement extends beyond the midline

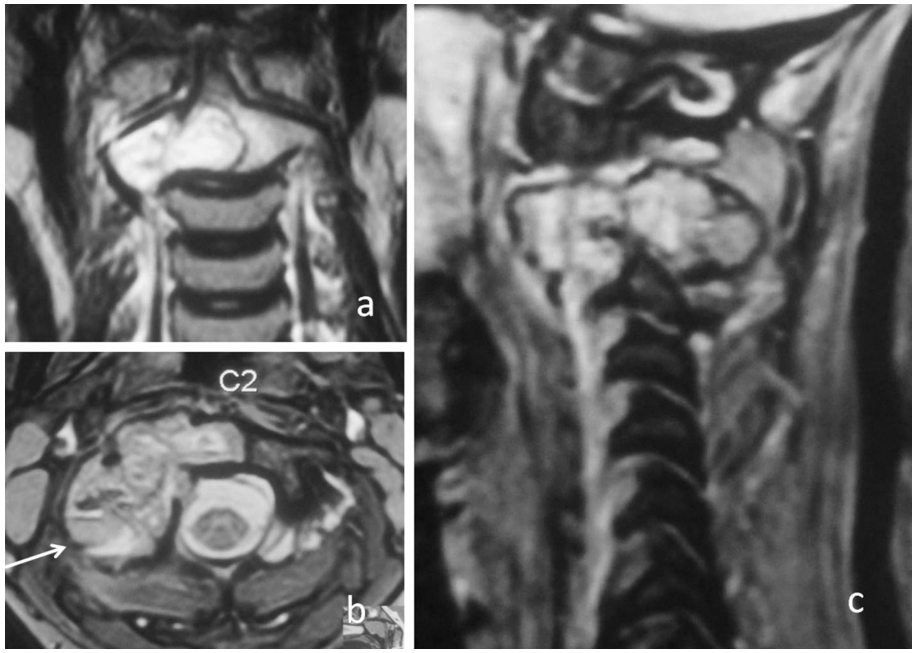

Fig. 2 Pre-operative MRI images. **a** Coronal section showing involvement of the right lateral mass and body extending up to midline. **b** The axial section with (*arrow*) depicting fluid levels. **c** Sagittal section shows the tumor extending into the posterior elements and involving the entire right facet of C2

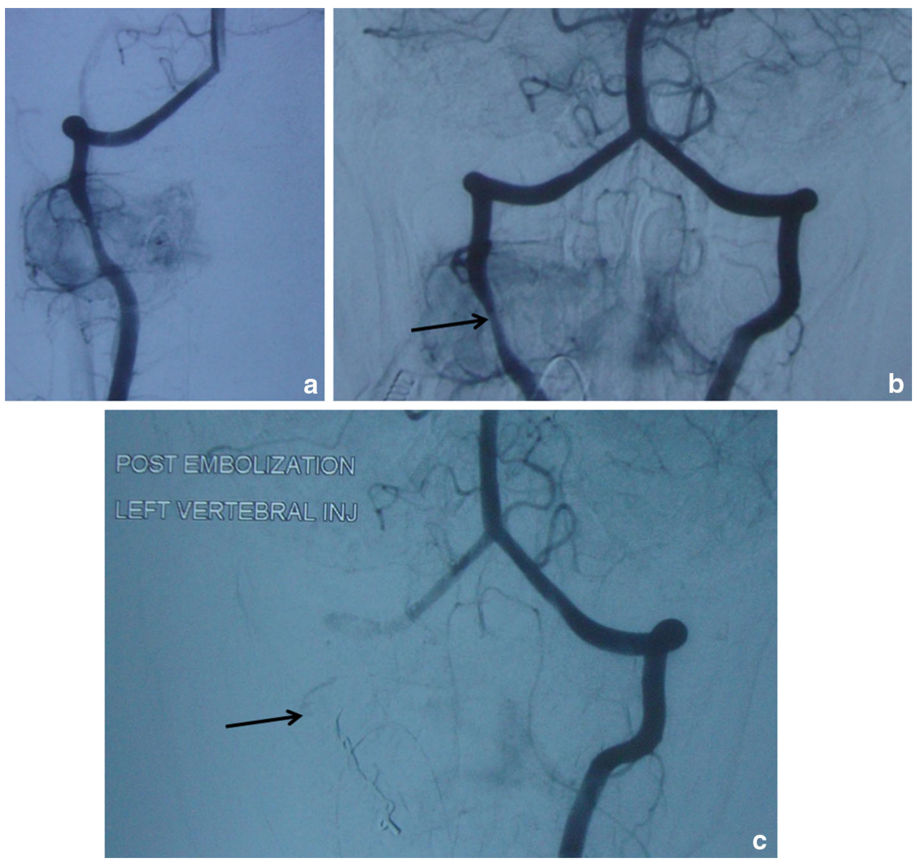

Fig. 3 Images of embolisation procedure. **a** Tumor blush noted encasing the vertebral artery with narrowing noted in the centre of the tumor. **b** Left vertebral well visualised with normal basilar flow and (*arrow*) depicting the stenosis of the vertebral artery. **c** Post embolisation images show reduced vascularity

ABC have classically been divided into primary and secondary types. Primary ABC arise de novo and form about 70 % of the cases. Secondary ABC arise from pre-exsisting lesions such as chondroblastoma, osteoblastoma and telangiectatic osteosarcoma and comprise of 20–30 % cases [4, 5]. There is an additional rare variant called the "solid" variant which is so named due to paucity of cavernous spaces in the tumor. This variant accounts for 3–8 % [4].

Diagnosis

Local pain is the most common presenting feature [4, 5, 7, 8]. The tumor may be associated with a discernable swelling which may show tenderness on palpation or result in deformity [9–12]. These tumors can cause significant local bony destruction resulting in compression of adjacent at risk structures, which include the spinal cord, nerve roots, vertebral artery in cases of cervical spine tumors and soft tissue structures in the immediate vicinity of the tumors. Neurological deficit may be seen at the time of presentation in 60–70 % of cases and vary from mild radiculopathies, myelopathy to overt neurological deficit [1, 4, 8, 11]. Diagnosis is confirmed with histopathological examination especially when primary bone lesion is suspected. Imaging modalities such as X-ray and CT scan show a lytic lesion with ballooning of the cortex, egg shell appearance often resulting in a breach of the cortex with pathological fracture [1, 4, 13]. MRI appears to be the gold standard for diagnosis. Fluid–fluid interface is a characteristic appearance for ABC on T2 weighted images. They have a heterogeneous appearance on T1 and T2 weighted images and septa show enhancement on administration of Gadolinium [13]. Selective angiography is a valuable tool and can help in identification feeder vessel, selective embolisation may be performed simultaneously to reduce vascularity and intraoperative blood loss at later surgery [7, 8].

Pathology

ABC is a benign cystic lesion which contained blood filled cystic cavities separated by connective tissue septa containing fibroblasts, osteoclast-type giant cells and reactive bone.

The exact pathogenesis of development of ABC has not been clearly elucidated [5]. It is postulated that ABC results from secondary vascular lesion developing in a primary bone lesion [4, 5]. The hemodynamic pressures for the arteriovenous channels cause erosion in the trabecular bone and cause formation of enlarged cystic cavities. The periosteal reaction initiates an osteoblastic and osteoclastic activity resulting in the classical ballooned appearance of the lesion. These blood filled cavities are not true cavernous lesion as they do not have an endothelial lining and smooth muscle which is seen in the walls of blood vessels.

Differential diagnosis

Differential diagnosis includes other causes of lytic lesions like hemangiomas, fibrous dysplasia, giant cell tumors and metastatic deposits [5]. Features which help in diagnosis include age of presentation, typical MRI findings, solitary lesion in the posterior elements, sparing of the disc space and pattern of bony destruction [1, 4, 5]. However, in case of atypical features histopathology provides the final diagnostic evidence.

Rationale of treatment

The treatment options used for treating cases of aneurysmal bone cysts vary based on a wide array of factors. The factors that need to be taken into consideration include site or location of tumor, age of the patient, neurological deficits, associated deformity, presence of pathological fractures and potential postoperative instability after complete resection.

Authors have suggested intralesional surgery to be adequate treatment for such tumors if lesion is small and does not extend into the paraspinal regions [3, 4]. They also suggested that selective arterial embolisation can be as effective as intralesional surgery and has the added benefit of being minimally invasive and cost effective for treatment for such cases [3, 7, 8]. However, cases which show pathological fracture, neurological deficit, scenarios not amenable to embolisation and cases which have reoccurred following previous embolisation should undergo surgical decompression, intralesional surgery and reconstruction following decompression [1, 3, 4, 7].

Radiotherapy also is effective treatment for such tumors, however, there is higher risk of post radiation deformity when accompanied by surgical curettage. [1].

Although radiation alone is effective in some cases Capanna and others noted higher rates of reoccurrences when used alone. This treatment modality has the risk of late malignancy in the irradiated tissue especially when used in the pediatric age group [2]. They suggested radiation only in situations where tumor location precluded surgical treatment options and as an adjuvant where patient has been left with residual tumor lesion after surgery [2]. There are occasional reports on the use of intralesional calcitonin and methylprednisolone in scenarios where surgery and embolisation are not feasible [14].

Surgery is the treatment of choice when stability of the spine is compromised and neurologic signs are present. Complete resection remains the treatment of choice, particularly for patients who present with neurological deficits and provides highest rate of cure [1–3, 8]. Incomplete excision is associated with a higher rate of recurrence of up to 60 % [3].

The growth of an aneurysmal bone cyst results in rapid enlargement and bone destruction, leading to neurological compromise. Total excision must include the entire cyst as well as the abnormal surrounding tissue and the bony surfaces that are lined with the hypervascular membrane. Intraoperative bleeding usually subsides when the cyst wall is removed [1]. ABCs have a large propensity to involve posterior elements. These tumors need a posterior surgical excision and any tumor extension anteriorly would benefit additional staged anterior tumor resection [1–3, 15]. Complete tumor excision would be ideal as reoccurrence rates with residual tumor are high. Large tumor growth can result in deformity and instability; surgical resection of such tumors may result in an iatrogenic instability [9, 10, 12]. Such scenarios should be dealt with reconstruction of the created defect with bone grafting and instrumented stabilization. Posterior instrumentation using lateral mass screws, pedicular screws or hooks and rods can be performed in the same instance. de Kleuver et al. emphasize the problems of a posterior approach alone in cases with anterior extension of tumor which seems to have a higher rate of recurrence [3]. Spinal stabilization would also be desirable with cases which involve junctional areas to avoid the risk of post laminectomy kyphosis and deformity [9, 10].

Rationale of authors' approach

The treatment options for aneurysmal bone cyst include a large number of non surgical options including selective arterial embolisation, use of sclerotherapy, intravenous bisphosphonates and intravenous Denosumab [16–21]. In fact there have also been reports of spontaneous resolution of ABC lesions following fracture and diagnostic biopsy. McQueen et al. and Malghem et al. noted spontaneous resolution of ABC lesions in the pelvis, femur and lumbar spine [22, 23]. Malghem reported on 3 cases aged 18, 19, and 22 years and concluded that an older age at presentation may lead to spontaneous resolution [23]. Recently Denosumab, a monoclonal antibody which blocks osteoclast function has shown benefit in treatment of giant cell tumors. Lange reported on the use of Denosumab in two cases of recurrent C5 ABC with satisfactory short-term results, however, they concluded that data pertaining to the use of Denosumab is still lacking in pediatric patients and longer follow-up studies may be needed to establish a definitive role of Denosumab in the treatment of ABC [21]. Results of selective arterial embolisation were documented by Amendola et al. in seven cases of spinal ABC lesion with satisfactory results with cases requiring repeated embolisation ranging from one to seven procedures [17]. They concluded that it was a cost effective treatment option especially in cases without pathological fracture and neurological deficit with an intact cyst wall. Rossi reported on over 400 embolisation procedures for various bone tumors and found embolisation to produce satisfactory results in treatment of ABC lesion with 94 % healing of lesion with repeated embolisation procedures [16, 18]. Dubois reviewed results of use of percutaneous sclerotherapy in the treatment of ABC in 17 cases with 2 cases involving the thoracic spine and sacrum respectively. They noted excellent results, however, nine of their cases needed repeated procedures [24]. Serial intravenous zolendronic acid infusion has documented healing response in giant cell tumors and was noted to be useful in the treatment of a sacral ABC but the authors concluded that larger study would be needed to observe the efficacy of this treatment modality in the treatment of ABCs [20].

In the case discussed involvement of the C2 vertebral precluded the use of a percutaneous sclerotherapy option. We felt that use of emerging pharmotherapeutic options including Denosumab and bispohosphonates though attractive still do not have adequate safety profile data and long-term outcomes pertaining to the use in pediatric population with ABCs.

We considered preoperative embolisation to reduce the vascularity of the tumor. The patient hailed from a remote part of the country needing considerable time and financial expenditure to travel to the institute which resulted in logistic limitation in seeking repeated intervention procedures. There was complete involvement of the right neural arch and lateral mass of the C2 vertebra with cortical breech and soft tissue extension. In view of the risk of instability and following a discussion of treatment options with the patients family surgical treatment option was planned.

The prime concerns in the preoperative planning of tumor resection for this case revolved around the following parameters. (a) feasibility of single stage procedure; (b) surgical approach to C2 to facilitate best possible tumor clearance; (c) need to provide a stable and good reconstruction of the defect following tumor clearance; and (d) possibility to preserve motion segments and avoiding extension of fixation to occiput.

Evaluation of the tumor as per Weinstein–Boriani–Biagini classification showed involvement of zones 2–8 with tissue layer extension from A to D thus involving a significant extent of both anterior and posterior columns to preclude the possibility of a single stage surgical excision [25, 26]. The best chance of complete tumor clearance could be obtained with combined anterior and posterior procedures. A first stage posterior surgery was preferred as a preliminary anterior procedure would create a large post

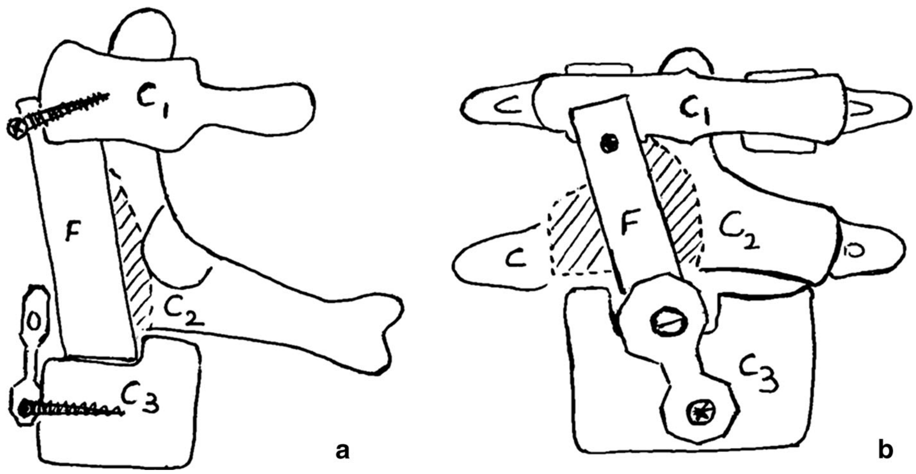

Fig. 4 Line diagram depicting the graft position and placement of instrumentation. **a** Lateral view showing the Fibula graft marked as 'F' fixed between the lateral mass of C1 and C3 in the void created following tumor excision (*area shaded*). **b** Depicting use of a buttress plate fixed to C3 to prevent graft dislodgment

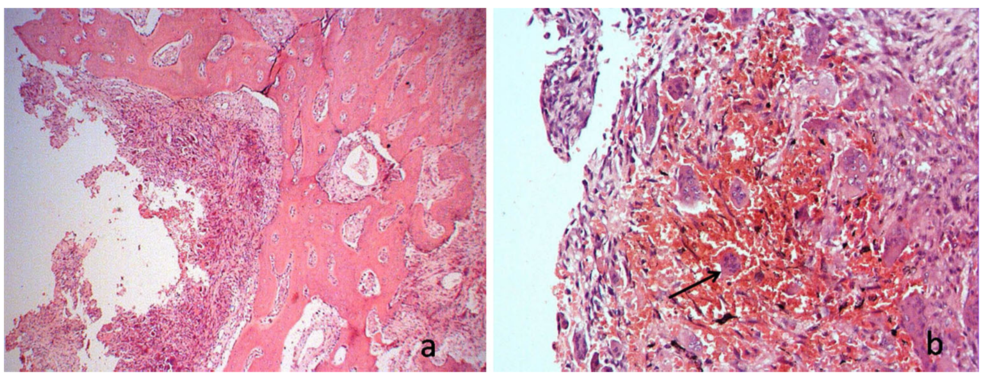

Fig. 5 Histopathology images of the tumor Hematoxylin and Eosin staining. **a** Low magnification image showing numerous blood filled spaces lined by giant cells and mononuclear cells at (×4). **b** High magnification image showing (*arrow*) depicting giant cells. There was no evidence of necrosis, mitosis or granulomatous lesion (×40)

tumor excision defect with significant instability and risk of neurological injury. An initial posterior approach tumor excision with simultaneous posterior instrumentation and stabilization was considered to be suitable and was the preferred option. Possible posterior instrumentation strategies included multilevel segmental fixation with screws including lateral mass and pedicular screws and sublaminar wiring techniques. The dimensions of the lateral mass on pre-operative CT scan planning were 5–6 mm and were considered to be inadequate to achieve satisfactory fixation using lateral mass screws. Sublaminar wires would require long segment fixation, sacrifice of significant number of motion segments and extension to the occiput to offer the best chance of success.

Therefore pedicular screw fixation was the preferred fixation strategy. Pedicular screw placement in the pediatric cervical spine especially in the subaxial spine at the C3, 4 is a significantly challenging proposition as evidenced by morphometric studies on pedicle dimensions [27]. Navigation assistance was considered necessary in this case to aid placement of pedicular screws in the subaxial spine. Our previous experience with use of Iso-C navigation system aided cervical pedicular screw placement has resulted in safe and accurate screw placements [28]. The supplementary option of preoperative angiography with embolisation was sought to reduce the intraoperative blood loss and to ascertain the integrity and dominance of the vertebral artery.

Anterior surgical approach to C1–2 includes the options of transoral approach, lateral retropharyngeal approach, modified anterior Southwick and Robinson approach and Mac Afee retropharyngeal approach. The other options of mandible splitting and extended maxillectomy approaches to gain access to C1–2 are substantially disfiguring and were not considered. Modified McAfee anterior retropharyngeal approach was preferred in this scenario since it provides excellent access to the C1–2 junction to allow complete tumor excision. This approach has the advantages of being retropharyngeal and the oral cavity is not entered. There is no dissection posterior to the carotid sheath and

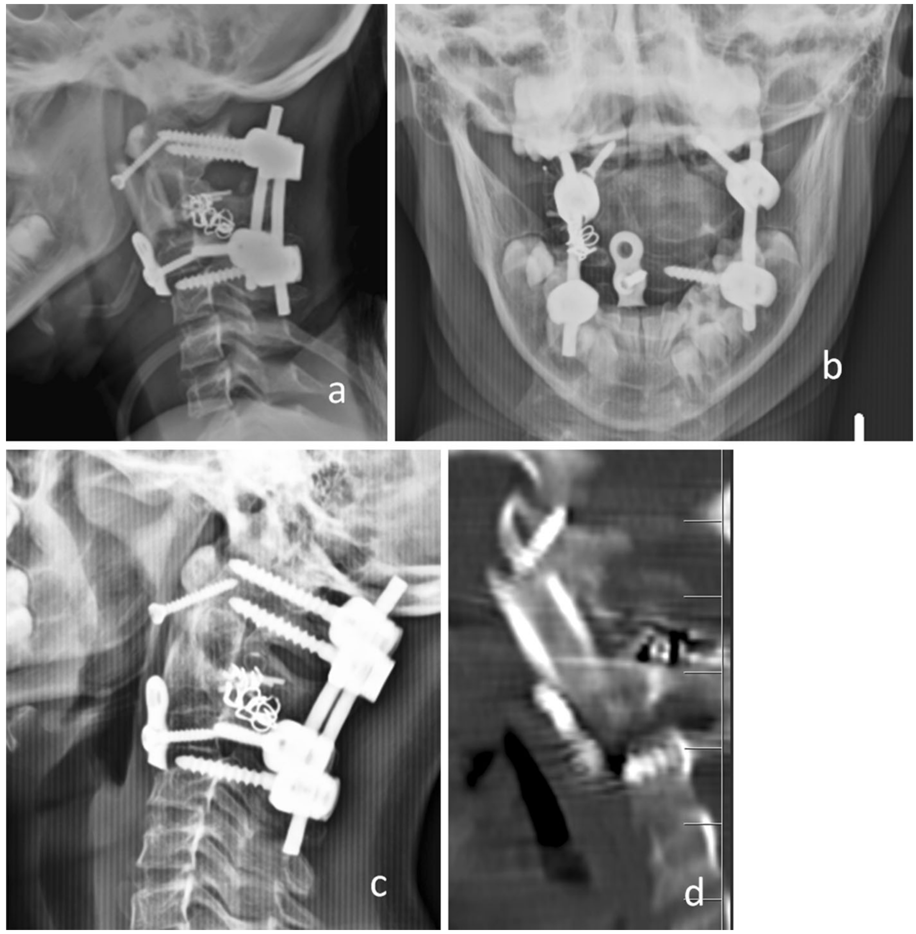

Fig. 6 Post-operative images. **a**, **b** Immediate post-operative X-ray showing the fibular graft spanning C1 lateral mass and C3. A buttress plate fixed to C3 to prevent graft extrusion. **c** X-ray at final follow-up at 12 months showing fusion. **d** CT scan done at 6 months with graft incorporation and maintained graft position

lower risk of post-operative airway obstruction compared to the lateral retropharyngeal approach. This approach allows adequate space to perform anterior satisfactory tumor clearance and reconstruction with a fibular strut graft which is cumbersome using a Southwick Robinsons approach especially at the C1–2 region.

The fibular strut graft was selected to provide stable anterior column reconstruction. To provide firm graft anchorage it was fixed to the C1 lateral mass with a screw and wedged into the defect following tumor excision. The lower graft anchorage was achieved with a 2 holed buttress reconstruction plate fixed to the anterior aspect of C3. This configuration provided a stable fixation with minimal chance of graft dislodgment (Fig. 4).

First stage

Under general anesthesia with Mayfield clamp applied, posterior approach with exposure from C1 to C4 was performed. Under navigation guidance bilateral C1 lateral mass screw, pedicle screws were inserted into C3 on the left and C4 on the right side.

A highly vascular and well encapsulated tumor was noted in the right lamina, lateral mass and pedicle intra-operatively. The tumor mass was excised from right C2 lateral mass in piece-meal fashion. The right vertebral artery was isolated and clamped by using vascular clamps to aid in maintaining hemostasis. For enhancing the surgical exposure of the tumor right C2 nerve root ganglion was divided after ligation. Access to the tumor tissue in the anterior elements including C2 vertebral body was achieved and tumor excision was performed. The posterior elements of C2, C3, C4 were decorticated and bone grafting using allograft was done over the prepared fusion bed. Contoured rods were connected to the pedicle screws and lateral mass screws and wound closed.

Second stage

Skin incision was made along right mandibular margin. Dissection was performed to isolate the submandibular gland medially, facial vein and digastric muscle supero-laterally and the carotid sheath with vessels laterally. The anterior C1–2 region was exposed. The thinned out cortex

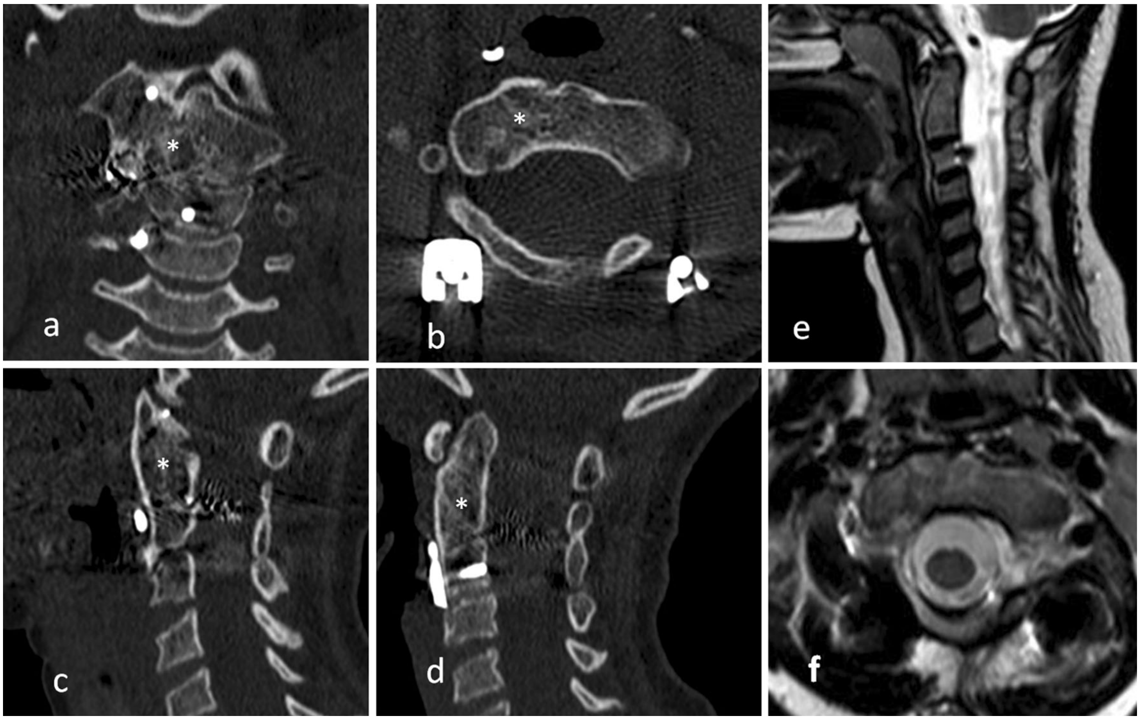

Fig. 7 **a–d** CT scan done at 27 month follow-up shows excellent graft incorporation (area of tumor marked with asterix) with fusion and no evidence of residual tumor. **e**, **f** T_2 weighted MRI images with no evidence of tumor recurrence

was broken and tumor cavity was exposed. Through clearance of remnant tumor tissue was completed and walls of the cavity were adequately curetted. Fibular strut graft was harvested and shaped into a size, appropriate to fill the post-tumor excision void. The superior and inferior end of the graft was fashioned to fit obliquely into the defect. The graft was snugly fit between the C1 lateral mass and the body of C3. Superior end of graft was fixed to right C1 lateral mass with 2.7 mm screw. Inferior end of graft was wedged into a slot in the C3 vertebral body and the entire configuration was stabilised with the help of 2 holed reconstruction buttress plate. Implant positioning was satisfactory on intra-operative fluoroscopy. Wound was closed. Postoperative period was uneventful. The patient remained neurologically intact and was discharged after 1 week and was advised to use a cervical orthosis in the form of a SOMI brace for 6 weeks.

Procedure imaging section

The histopathological examination of the excised specimen revealed heterogenous lesion with blood filled spaces lined by giant cells and mononuclear cells (Fig. 5). The fibrous septa were made of fibroblasts, myofibroblasts, hemosiderin deposits, blood vessels, and fields of osteiod and woven bone. The cells were arranged in solid sheets with focal cystic formation. Osteoclast-like giant cells were interspersed within the more solid areas. The periphery of the lesion displayed a shell of benign reactive bone forming a capsule. There was no evidence of granuloma formation or necrosis. There was no evidence to suggest a giant cell tumor or fibrous dysplasia or malignancy. These features were compatible with the diagnosis of an aneurysmal bone cyst.

The immediate post-operative X-ray and CT scan showed satisfactory implant position and good position of the graft (Fig. 6).

Outcomes

Follow-up examination at 6 weeks showed complete relief from pain and neurology was normal. Follow-up radiographs showed good position of implants (Fig. 6). At last follow-up at 27 months patient was asymptomatic and doing well with radiological fusion evident on plain radiographs and CT scan; the follow-up MRI did not show any evidence of tumor recurrence (Fig. 7).

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Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

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