Conditions resulting in atlantoaxial dislocations (AADs) are rare but have the potential for severe consequences of neurological deterioration, such as myelopathy, paralysis, respiratory failure, or even death.\(^8\)\(^{,}\)\(^{20}\) AAD typically requires surgical treatment with several atlantoaxial fixation and fusion methods.\(^{18}\)\(^{,}\)\(^{20}\) The available techniques of posterior atlantoaxial instrumentation include pedicle screw (PS) fixation at C-2, C-1 and C-2 transarticular screws (TASs), C-2 laminar screws (LSs), and lateral mass screws (LMSs) at C-1, among others.\(^{20}\) The connection in fixation methods can be achieved using two general methods: rod or plate designs.\(^{4}\)\(^{,}\)\(^{5}\) Cervical fixation methods are often bilateral and symmetric; however, concomitant osseous or vascular deformities at the craniovertebral junction can result in hypaxial dislocations among patients with cervical osseous or vascular abnormalities utilizing hybrid techniques

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OBJECTIVE Most cervical fixations for atlantoaxial dislocation (AAD) are bilateral and symmetric; however, in the setting of osseous and vascular deformity at the craniovertebral junction, asymmetrical and hybrid fixations are used as “salvage” techniques. Because of the rarity of these cases, hybrid cervical fixations for AAD have not been fully explored. The aim of this study was to evaluate the clinical feasibility and outcomes of posterior hybrid cervical fixations for AAD.

METHODS Twenty-one AAD cases were retrospectively studied; 18 had cervical myelopathy with Japanese Orthopaedic Association (JOA) scores ranging from 9 to 16 (mean 13.5). Hybrid fixation techniques included unilateral pedicle screws, transarticular screws, C-2 laminar screws, cervical lateral mass screws, and spinous process screws. During the same period, 82 AAD cases, treated using symmetric traditional fixations, were analyzed as controls.

RESULTS Atlantoaxial fixation was performed in 11 cases, while occiput-cervical fixation was used in 10 cases. All cases achieved solid osseous fusion. Anatomical reduction was achieved in 20 cases (95.2%). All 18 cases with myelopathy showed postoperative improvement, with JOA scores ranging from 13 to 17 (mean 15.5). Three cases (14.2%) experienced complications, including delayed wound healing, CSF leakage, and fixation loosening. Hybrid fixation techniques showed significantly greater estimated blood loss when compared with controls (208.1 ± 19.30 ml vs 139.63 ± 8.75 ml, \(p = 0.001\)). Operative duration (125.38 ± 6.29 min vs 119.41 ± 3.77 min, \(p = 0.464\)), complication rates (14.3% vs 4.9%, \(p = 0.148\)), and JOA improvement rates (61% ± 7% vs 49% ± 4%, \(p = 0.161\)) showed no significant differences.

CONCLUSIONS For AAD with osseous or vascular deformity, posterior cervical reduction and stabilization can be achieved using hybrid techniques, resulting in comparable clinical results to symmetric traditional fixation.
niovertebral junction prohibit symmetrical fixation techniques. Thus, asymmetrical hybrid fixations are frequently used as “salvage” procedures.9,14

Several authors previously reported on alternative or salvage C-2 LS placement in lieu of traditional fixation techniques, such as PS fixation;2,6 because of the rarity of these cases, the feasibility and results of salvage hybrid cervical fixations for AADs have not been fully explored. In the current series, the clinical features, treatment pitfalls, and surgical outcomes of 21 cases are investigated.

Methods
Study Design and Clinical Features

This study was a retrospective review of a prospectively collected database. IRB approval from Peking University Third Hospital and informed patient consent were obtained prior to study enrollment. One hundred and three patients with AAD were treated surgically by the senior author (S.W.) from September 2008 to September 2013. The sole criteria for the presence of AAD was an atlantodental interval (ADI) greater than 3 mm; for patients younger than 18 years of age, greater than 5 mm was used. Among these 103 AAD cases, 21 patients with upper cervical osseous or vascular deformities underwent surgical reduction and fixation utilizing hybrid atlantoaxial fixations. Hybrid fixation was defined as the use of two or more asymmetrical fixation techniques. Analysis was retrospectively performed in this series. The mean follow-up duration was 45.0 months (range 36–60 months). There were 14 females and 7 males included. The ages ranged from 7 to 71 years, with a mean of 44.2 years. Fourteen cases were classified as reducible AAD, while 7 were determined to be fixed AADs. Eighteen cases had myelopathy on presentation (Japanese Orthopaedic Association [JOA] score range 9–16, mean 13.5). Prior to surgical correction, the patients underwent dynamic lateral radiographs, as well as reconstructive CT and MR imaging of the cervical spine. Among the 21 cases, 9 had evidence of occipitalization of the atlas, 6 had Klippel-Feil syndrome (KFS) with C-2 and C-3 congenital fusions, and 1 had a C-2 to C-6 congenital fusion (KFS). Four cases had tonsillar herniations (Chiari malformation type I deformities) and 3 had syringomyelia. Further details of radiographic findings and clinical data are presented in Table 1. The other 82 cases with symmetric traditional fixations were analyzed as the control group. The control cases underwent surgery by the same surgeon, and the intraoperative and postoperative procedures (except for the fixation techniques) were kept consistent with the “hybrid” cases.

Surgical Methods
Instrumentation and Fusion Length (fixed and fused segmentation)

To preserve cervical mobility, the shortest instrumentation length was preferred; thus, atlantoaxial fixation was always the first option. In hypoplasia or occipitalization of the atlas, fixation extending from the occiput to C-2 was used. In KFS cases, the fixation was typically extended only to the congenital fused segments, without further sacrificing the lower cervical movements.

Determination of Initial Technique and Salvage Procedures

C-1 and C-2 PSs or TASs were always the initial option for fixation, because of the solid biomechanical features. In the setting of a “high-riding” vertebral artery (VA) at C-2, or C-2 PS insertion failure, C-2 LSs were chosen. For patients with KFS, the fixation was extended to the subaxial spine, preferably with cervical PS placement at these caudal levels. LMSs were used alternatively for lower cervical pedicle hypogenesis. We found that connecting C-2 LSs with lower LMSs was challenging and required acute rod angulation. To strengthen C-2 LSs in KFS, cervical spinous process screws (SPSs) were used. Only cases undergoing asymmetrical fixations were retrospectively analyzed.

Lateral Mass, PS, and Plate Fixation of C-1 and C-2.

The C-1 (usual diameter 3.5 mm, length 30 mm) and C-2 PS (diameter 3.5 mm, length 26–28 mm) was inserted, followed by placement of the reconstruction plate between C-1 and C-2 (Fig. 1).

Transarticular C1–2 Fixation (modified Magerl’s technique without the cable). A 3.5-mm full-threaded cortical titanium screw (length 38–42 mm) was inserted across the C1–C2 lateral mass joint. We did not use cable in this study.15

Occipit to C-2 Fixation Using C-2 PSs. Patients with occipitalization of the atlas or dysplasia of the C-1 posterior arch underwent this technique. C-2 PSs were placed as previously described. The occiput and C-2 were connected by a reconstruction plate. Three 3.5-mm bicortical screws were placed into the occiput.

C1–2 or Occiput to C-2 Fixation Using C-2 LSs. Among patients with VAs that were high-riding (Fig. 1) or in patients with osseous anomalies of the C-2 pedicle, C-2 LSs were used as a salvage technique for C1–2 stabilization, or in select cases of occipitocervical fixation. The procedure used the technique of Wright.19 The LS was connected with C-1 or occipital screws using a titanium rod (Fig. 1).

Cervical LMS. In KFS cases, the lower cervical LMSs (diameter 3.5 mm, length 12–16 mm) were connected with a C-2 PS, strengthening the hybrid fixation.

Lower Cervical SPSs. An entry point was created with a burr at the bottom of the spinous process. The screw trajectory was drilled toward the contralateral laminar surface and penetrated out from the junction of the lamina and the spinous process, and without penetrating into the lamina. After checking and tapping, the screws were fixed on the spinous process bicortically. The length of the SPSs in this report ranged from 12 to 14 mm. Then the screws were connected with the C-2 LSs with rods.

Procedures After Fixation

At the end of the procedure, for all 103 cases, the C-1 arch, C-2 lamina, and spinous process were decorticated with a high-speed burr. Morselized cancellous grafts harvested from the posterior iliac crest were bridged between the C-1 arch and C-2 lamina. For 100 cases, no brace was used postoperatively. However, in 3 “hybrid” cases, severe dislocation and viscoelastic rebound forces were
<table>
<thead>
<tr>
<th>Case No.</th>
<th>Age (yrs), Sex</th>
<th>Diagnosis</th>
<th>Hybrid Categories &amp; Fixation Segments</th>
<th>Reasons for Using Salvage Techniques</th>
<th>Complications</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>52, M</td>
<td>Reducible AAD, OO, myelopathy</td>
<td>2 (C-2 PS + C-2 LS), C1–2 fixation</td>
<td>High-riding VA at C-2 segment</td>
<td>Iliac wound exudation &amp; delayed healing</td>
</tr>
<tr>
<td>2</td>
<td>47, F</td>
<td>Irreducible AAD, occipitalization, BI, TH, myelopathy</td>
<td>2 (C-2 PS + C-2 LS), C1–2 fixation</td>
<td>High-riding VA at C-2 segment</td>
<td>NA</td>
</tr>
<tr>
<td>3</td>
<td>44, F</td>
<td>Irreducible AAD, myelopathy, occipitalization, KFS (fused C2–3/C5–6)</td>
<td>2 (C-2 PS + C-2 LS), Oc–C2 fixation</td>
<td>Blocked &amp; hypoplastic C-2 rt pedicle in KFS case</td>
<td>NA</td>
</tr>
<tr>
<td>4</td>
<td>58, M</td>
<td>Reducible AAD, myelopathy</td>
<td>2 (C-2 PS + C-2 LS), C1–2 fixation</td>
<td>High-riding VA at C-2 segment</td>
<td>NA</td>
</tr>
<tr>
<td>5</td>
<td>38, M</td>
<td>Reducible AAD, OO</td>
<td>2 (C-2 PS + C-2 LS), C1–2 fixation</td>
<td>High-riding VA at C-2 segment</td>
<td>NA</td>
</tr>
<tr>
<td>6</td>
<td>39, F</td>
<td>Reducible AAD, OO, myelopathy</td>
<td>2 (C-2 PS + C-2 LS), C1–2 fixation</td>
<td>High-riding VA at C-2 segment</td>
<td>NA</td>
</tr>
<tr>
<td>7</td>
<td>38, F</td>
<td>Irreducible AAD, occipitalization, myelopathy, BI, hypoplastic laminae of C-2 &amp; C-3</td>
<td>2 (C-2 PS + C-3 LS), Oc–C3 fixation, &amp; halo vest for 3 mos</td>
<td>Hypoplastic C-2 laminae w/hypotrophic pedicle of C-2 (rt side)</td>
<td>NA</td>
</tr>
<tr>
<td>8</td>
<td>31, F</td>
<td>Irreducible AAD, myelopathy, BI, KFS (fused C2–3)</td>
<td>2 (C-2 PS + C-2 LS), C1–2 fixation</td>
<td>Blocked &amp; hypoplastic C-2 rt pedicle in KFS case</td>
<td>NA</td>
</tr>
<tr>
<td>9</td>
<td>61, F</td>
<td>Irreducible AAD, OO</td>
<td>2 (C-2 PS + C-2 LS), C1–2 fixation</td>
<td>High-riding VA at C-2 segment</td>
<td>NA</td>
</tr>
<tr>
<td>10</td>
<td>61, M</td>
<td>Reducible AAD, old odontoid fracture, myelopathy</td>
<td>2 (C-2 PS + C-2 LS), C1–2 fixation</td>
<td>High-riding VA at C-2 segment</td>
<td>NA</td>
</tr>
<tr>
<td>11</td>
<td>70, M</td>
<td>Reducible AAD, OO, myelopathy, Kaschin-Beck disease</td>
<td>2 (C-2 PS + C-2 LS), C1–2 fixation</td>
<td>High-riding VA at C-2 segment</td>
<td>NA</td>
</tr>
<tr>
<td>12</td>
<td>19, M</td>
<td>Irreducible AAD, occipitalization, BI, myelopathy, syringomyelia, torticollis</td>
<td>2 (C-2 PS + C-2 LS), Oc–C2 fixation, &amp; halo vest for 4 mos</td>
<td>Blocked &amp; hypoplastic C-2 rt pedicle in KFS case</td>
<td>Screws loosening &amp; dislocation recurred at 4 mos postop, &amp; performing transoral revision</td>
</tr>
<tr>
<td>13</td>
<td>50, F</td>
<td>Reducible AAD, OO, myelopathy</td>
<td>2 (C-2 PS + C-2 LS), C1–2 fixation</td>
<td>High-riding VA at C-2 segment</td>
<td>NA</td>
</tr>
<tr>
<td>14</td>
<td>12, F</td>
<td>Irreducible AAD, myelopathy, BI, occipitalization, KFS (fused C2–3), TH, syringomyelia</td>
<td>2 (C-2 PS + C-2 LS), Oc–C2 fixation</td>
<td>Blocked &amp; hypoplastic C-2 rt pedicle in KFS case</td>
<td>NA</td>
</tr>
<tr>
<td>15</td>
<td>7, M</td>
<td>Reducible AAD, myelopathy, vertebral hypogenesis of C2–6</td>
<td>2 (C-2 PS + C-2 LS), Oc–C2 fixation, &amp; halo vest for 3 mos</td>
<td>Vertebral hypogenesis of C2–6 &amp; C-2 PS plantation failure</td>
<td>NA</td>
</tr>
<tr>
<td>16</td>
<td>71, F</td>
<td>Reducible AAD, occipitalization, myelopathy</td>
<td>2 (C-2 PS + C-2 LS), Oc–C2 fixation</td>
<td>High-riding VA at C-2 segment</td>
<td>NA</td>
</tr>
<tr>
<td>17</td>
<td>17, F</td>
<td>Reducible AAD, occipitalization, myelopathy, KFS (fused C2–3), TH</td>
<td>3 (C-2 PS + C-2 LS + C-3 LMS), Oc–C3 fixation</td>
<td>Blocked &amp; hypoplastic C-2 pedicles in KFS case, &amp; high-riding VA at C-2 segment</td>
<td>NA</td>
</tr>
<tr>
<td>18</td>
<td>56, F</td>
<td>Reducible AAD, OO</td>
<td>2 (C-2 PS + C-2 LS), C1–2 fixation</td>
<td>High-riding VA at C-2 segment</td>
<td>NA</td>
</tr>
<tr>
<td>19</td>
<td>50, F</td>
<td>Reducible AAD, occipitalization, KFS (fused C2–3), myelopathy, swallowing disturbance</td>
<td>4 (C-2 PS + C-2 LS + C-3 LMS + C-3 SPS), Oc–C3 fixation</td>
<td>Blocked &amp; hypoplastic C-2 pedicles in KFS case</td>
<td>CSF leakage was treated successfully by prolonged drainage &amp; pressure dressing</td>
</tr>
<tr>
<td>20</td>
<td>59, F</td>
<td>Reducible AAD, C-1 hypogenesis, KFS (fused C2–6), myelopathy</td>
<td>3 (C-2 LS + C-2/3/6 LMS + C-3/5 SPS), Oc–C6 fixation</td>
<td>Blocked &amp; hypoplastic C-2–6 pedicles in KFS case, &amp; C-1 screw plantation failure</td>
<td>NA</td>
</tr>
<tr>
<td>21</td>
<td>49, F</td>
<td>Reducible AAD, occipitalization, KFS (fused C2–3), myelopathy, TH, syringomyelia, articulation disorder</td>
<td>3 (TAS + C-2 PS + C-3 LMS), Oc–C3 fixation</td>
<td>Blocked &amp; hypoplastic C-2 pedicles in KFS case (C-2 PS partly cutting)</td>
<td>NA</td>
</tr>
</tbody>
</table>

BI = basilar invagination; NA = not applicable; OO = os odontoideum; TH = tonsillar herniation.
noted during the procedure, and additionally a halo vest was used postoperatively for supplemental stability (for 4 months after the fusion was confirmed). Drains were removed at 48 hours. After these procedures, patients were mobilized out of bed and were encouraged to participate in basic physical therapy. All 103 patients underwent postoperative radiographs and reconstructive CT scans of the cervical spine at 5 days and 4 months after the surgery. Radiographic examination was repeated at 12 months and annually thereafter. Fusion and screw position were confirmed by postoperative reconstructive CT (if fusion could not be confirmed using 4-month CT, we repeated reconstructive CT scans after 3 months). We used the Tan et al. criteria for assessing fusion, documenting bridging osseous union between the proximal and distal end point, which was determined to be grade 1. Density of volume of fusion was not assessed in this series.

Results
Among the 21 cases, 17 underwent posterior hybrid fixations using two distinct techniques, 3 used three techniques, while 1 case involved four distinct techniques. The reasons for hybrid fixations included: VA was high-riding at the C-2 segment, pedicle or vertebral hypogenesis at C-2, and blocked and hypoplastic pedicles in KFS cases.
Illustrative Case

This case (case 19) involved a 50-year-old woman with a 1-year history of progressive weakness and numbness of her lower extremities, and dysphagia. Neurological examination showed symmetrical hyperreflexia in the lower extremities. Hoffmann and Babinski signs were present bilaterally. Limited range of motion of the cervical spine on both extension and flexion radiographs was observed. Radiographs demonstrated a reducible AAD, occipitization of the atlas, and C2–3 congenital fusion (KFS). Reconstructive CT revealed that the left C-2 pedicle was well healed for 12 months and entered the C-2 vertebrae. The patient experienced good neurological recovery. Because the fixation was still rigid (as assessed by CT and dynamic radiography), we did not perform a revision.

The cases performed with hybrid techniques showed no significant differences compared with the control cases. In the 82 control cases, 4 patients suffered complications. One patient suffered incisional delayed healing (3 weeks), 1 suffered postoperative dysphagia for 6 months, 1 had severe occipital pain (lasting 3 months), and the last had a CSF leakage due to malpositioning of the halo vest. For the last case, we carefully observed him for more than 2 years and found no further complications. For the last case, we carefully observed him for more than 2 years and found no further complications.

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left side, C-2 PSs and C-3 LMSs were placed and connected with the occiput by a plate. Tightening of the cap on the C-2 screw achieved the atlantoaxial reduction. On the right side, a C-2 LS was used to avoid a VA injury. To strengthen the C-2 LS, a C-3 SPS was inserted and connected by a rod (Table 3). Morselized cancellous autograft harvested from the posterior iliac crest was bridged between the occiput and C-2 lamina. The postoperative CT scan showed complete reduction of the atlantoaxial joint, and the 4 used techniques were C-2 PS, C-2 LS, C-3 LMS, and C-3 SPS (Fig. 3H–K). The patient suffered postoperative CSF leakage (recognized during the drilling for occiput screws), which was treated successfully by prolonged drainage (72 hours) and pressure dressing. Postoperative CT (after 4 months) confirmed complete reduction and rigid fusion between the occiput and C-2. At the last follow-up at 2 years, the weakness of her lower extremities showed partial recovery (JOA score progressed from 14 to 16), and the dysphagia completely recovered.

Discussion

AAD treatment aims to stabilize the atlantoaxial joint with fixation and fusion without sacrificing the atlantooccipital and subaxial cervical joints. In the setting of occipitalization, fixation extending to the occiput does not additionally reduce the cervical collective movement; thus, fixation with extension to Oc–C2 fixation is universally recommended. For AAD with concomitant KFS (congenital cervical vertebra fusion), caudally extending the fixation to strengthen the instrument is reliable but should be kept within the congenitally fused segments.

In this report, fixation of all cases followed this principle. We have found that the screw-plate construct was fitted to reduce atlantoaxial displacement, especially for severe atlantoaxial anterior displacements. Thus, C1–2 PS fixation was the first option, and a screw-plate construct was preferentially used. Even in the setting of osseous deformities of the pedicle, a unilateral PS and plate construct for atlantoaxial reduction and fixation was used to obtain optimal results.

In the setting of a high-riding VA at the C-2 segment or hypogenesis of the C-2 pedicle (Table 3), the C-2 PS could not be inserted safely. To avoid VA injury, Wright described C-2 LSs in 2004, which have been widely used with favorable results. Because the axis of the C-2 LS
was different than that of the C-1 PS and the atlantoaxial displacement, we found the C-2 LS was a suboptimal anchor point for atlantoaxial reduction. We used unilateral PSs as a reductive anchor, and after the reduction, C-2 LSs were employed on the contralateral side to strengthen the fixation. Because of the large proportion of cancellous bone in the C-2 spinous process and lamina, the C1–2 construct of the C-2 LS was less rigid than that of the C-2 PS. Lapsiwala et al. found that C1–2 fixation with C-2 LSs had less stiffness in resisting lateral bending. We found the construct of C-1 PS with C-2 LS could passably bear the stress of atlantoaxial reduction. However, the stiffness

FIG. 3. Case 19. The dynamic radiographs of a 50-year-old woman showed reducible AAD, C-1 occipitalization, and C2–3 congenital fusion (A and B). Preoperative CT revealed the left C-2 pedicle could accommodate a 3.5-mm screw (C), while the right pedicle was occupied by the VA (D, arrow). The odontoid invaded the foramen magnum, and the ADI was abnormal (E). On the left side, C-2 PS and C-3 LMS were placed and connected with the occiput by a plate (F). On the right side, a C-2 LS and C-3 SPS were inserted and connected by a rod (F and G). Morselized cancellous grafts harvested from the posterior iliac crest were bridged between the occiput and C-2 lamina (G). The postoperative parasagittal CT scan showed the C-2 LS and C-3 LMS (H, arrow). The postoperative CT scan showed complete reduction of the atlantoaxial joint (I), and the 4 techniques included C-2 PS, C-2 LS, C-3 LMS, and C-3 SPS (J and K). Postoperative CT (after 4 months) confirmed complete reduction and rigid fusion between the occiput and C-2 (L, arrow). Figure is available in color online only.
of the occipitocervical fixation using C-2 LSs weakened, probably due to the longer lever arm and the occipitocervical rod possessing a more acute bend (from the failure of case 12). This hypothesis is supported by the work of Finn et al.\textsuperscript{3} Therefore, for KFS patients with the occipitocervical fixation using C-2 LS fixation, we recommend extending the fixation to C-3 and using additional halo vest stabilization.

In this report, the subaxial LMS and SPS were short (12–14 mm) and were performed as supplements to the PS or LS. The PS was easily connected with the LMS, while the LS connected with the SPS (Fig. 3). For patients with KFS, extending fixation to the congenital fused segment will not sacrifice the lower cervical movements. In this report, 3 SPSSs were used in 2 patients (at C-3 and C-5, respectively). Shin et al. evaluated the anatomical feasibility of LS placement in C3–7 using 3D screw trajectory software and found few patients could accommodate subaxial LS (13.5% at C-3, 19% at C-4, 0.9% at C-5, and 8.8% at C-6).\textsuperscript{10} Thus, the SPSSs at the subaxial cervical spine were used to strengthen C-2 LSs.

Limitations of the Study

We acknowledge several limitations to this study. First, the fact that the operating surgeon was one of two surgeons performing the clinical assessment and determining fusion status could potentially introduce bias into the results. However, these results still support the usage of alternative fixation techniques using the largest series to date of patients with AAD. Future investigations with blinded fusion assessment could help confirm the results presented here. In addition, follow-up duration remains limited at 45 months and further long-term follow-up data are required to assess the feasibility of the hybrid techniques.

Conclusions

The result of this largest series to date concerning usage of alternative fixation techniques in the setting of osseous or vascular deformities shows hybrid salvage fixation is a reasonable and favorable option for treatment of AAD. However, asymmetrical hybrid fixation is only recommended as a salvage technique because the fixation is significantly less robust than traditional techniques. In cases of severe atlantoaxial displacement or excessive superior odontoid migration, even though the reduction can be achieved, the weaker hybrid fixation experiences additional stress and may compromise outcomes. Thus, the excessive stress loading on the fixation needs to be considered. The performance of a transoral release as well as the addition of a halo vest is recommended to decrease the loading on the construct.

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References


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