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CLINICAL REPORT

Significance of prevention and early treatment of a postoperative twisted neck: atlantoaxial rotatory subluxation after head and neck surgery

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Abstract Atlantoaxial rotatory subluxation (AARS) is an infrequent condition that occurs most commonly in children for unknown reasons. Pediatric surgery, otopharyngeal inflammation, general anesthesia, and extreme rotation of the head are risk factors for development of postsurgical AARS, but AARS can often occur unnoticed, and the syndrome is not well known. We encountered three cases of postoperative AARS that occurred within 7 months; therefore, we have developed guidelines for prevention and early treatment of postoperative AARS. Postoperative AARS cannot be eliminated completely, but informed consent, a preoperative check, an appropriate surgical position, and a postoperative check may reduce the risk and damage related to this condition.

Keywords Preanesthetic assessment · Torticollis · Neck position · Postoperative recovery

Introduction

Atlantoaxial rotatory subluxation (AARS) is an infrequent condition that occurs most commonly in children [1–4]. Patients with AARS have torticollis and restricted neck motion with or without pain, which occur for unknown reasons after minor injury or upper airway infection. The presence of acquired torticollis in a child should arouse suspicion of AARS, especially during recovery from surgery performed on the head and neck under general anesthesia [2, 5], because 20–40% of cases of AARS are associated with surgery [3, 6–8]. This complication may be linked to the surgical position, but AARS can often occur unnoticed and is not well known, despite several case reports in the anesthetic literature [9–11]. As we encountered three cases of postoperative AARS that occurred within 7 months, we therefore have developed guidelines for prevention and early treatment of postoperative AARS.

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Case 1

The patient was a 7-year-old boy (120 cm, 21 kg) who was scheduled to undergo keloplasty for resection of a nevus sebaceous of the right ear. General anesthesia was induced with sevoflurane inhalation, and a classic laryngeal mask airway (LMA) was inserted without use of a neuromuscular blocker. Anesthesia was maintained with sevoflurane, oxygen, and nitrous oxide, again without a neuromuscular blocker. Pentazocine and an acetaminophen suppository were used for postoperative analgesia. Surgery was performed in the supine position with the head rotated laterally to the left. The surgical position was determined by the surgeon after induction of anesthesia, and the same position was maintained during surgery. The duration of surgery was 1 h 36 min. There was no finding when the neck was rotated back after surgery by the anesthetist.

Left torticollis with pain developed on postoperative day 1, and the range of motion of the neck decreased, but the patient was discharged on day 2. His torticollis continued, and a diagnosis of AARS was made based on computed tomography (CT) on day 5. The patient was admitted to our hospital for traction therapy. Improvement of AARS was seen on CT on day 10, and the patient recovered

completely after immobilization with a Philadelphia collar for 3 months.

Case 2

The patient was an 8-year-old girl (129 cm, 22 kg) with left cholesteatoma otitis who was scheduled to undergo tympanoplasty. General anesthesia was induced with midazolam followed by fentanyl and vecuronium. The trachea was intubated with a normal tube, and anesthesia was maintained with sevoflurane, oxygen, and nitrous oxide, in addition to vecuronium. Fentanyl was used for postoperative analgesia. Surgery was performed in the supine position with the head rotated laterally to the right. The surgical position was determined by the surgeon after induction of anesthesia, and the same position was maintained during surgery. The duration of surgery was 2 h 48 min. There was no finding when the neck was rotated back after surgery by the anesthetist.

Left torticollis with pain developed on postoperative day 1, and sternomastoid muscle spasm occurred on the left side, the contralateral side of the chin. The patient was examined by an orthopedic surgeon on day 5 and was diagnosed as myotonic torticollis. Outpatient traction therapy was applied, and the range of motion of the neck recovered, although residual torticollis remained.

Eight months later, tympanoplasty was performed to remove the residual cholesteatoma. General anesthesia was induced using sevoflurane inhalation followed by vecuronium. The trachea was intubated with a Ring–Adair–Elwyn (RAE) tube, and anesthesia was maintained with sevoflurane, oxygen, and air, in addition to remifentanil and vecuronium. Fentanyl was used for postoperative analgesia. Surgery was performed in the supine position with the head rotated laterally to the right. The surgical position was determined by the surgeon after induction of anesthesia, and the same position was maintained during surgery. The duration of surgery was 1 h 32 min. There was no finding when the neck was rotated back to neutral after surgery by the anesthetist.

Trismus with tonic neck and radiating pain appeared on postoperative day 1, and a diagnosis of AARS was made based on CT on day 11. Subluxation and reduction then occurred repeatedly, despite traction therapy. After 3 months of traction, the patient was treated surgically with posterior arthrodesis of C1–C2, and the range of motion of the neck is almost complete after 1 year.

Case 3

The patient was a 28-year-old woman (165 cm, 47 kg) who was scheduled to undergo surgery for a right cryptotia. General anesthesia was induced with propofol followed by fentanyl and vecuronium. The trachea was intubated with a normal tube, and anesthesia was maintained with sevoflurane, oxygen, and air. Fentanyl and flurbiprofen were used for postoperative analgesia. Surgery was performed in the supine position with the head rotated laterally to the left. The surgical position was determined by the surgeon after induction of anesthesia, and the same position was maintained during surgery. The duration of surgery was 2 h 23 min. There was no finding when the neck was rotated back to neutral position after surgery by the anesthetist.

The patient complained that she had difficulty in turning her head to the right on postoperative day 1 and received treatment with an antiinflammatory analgesic plaster. Right tonic neck and left torticollis developed, and AARS was diagnosed based on CT on day 2. Traction therapy was applied, and an improvement was observed on CT on day 6. The patient was discharged on day 11.

Discussion

A recent systematic review of the literature showed that 48% of cases of AARS (Grisel's syndrome) were caused by infection; 40% occurred postoperatively [6]. Of the cases caused by infection, upper respiratory tract infection was present in 83%, and of the postoperative cases, 78% occurred after adenotonsillectomy.

A possible pathway for spread of inflammation to the atlantoaxial ligaments has been demonstrated via direct anastomoses between lymphatic vessels and pharyngovertebral veins, and resultant instability may ensue after hyperemia as a consequence of the inflammatory process, decalcification of the vertebrae, and pathological relaxation of the transverse ligament [4, 9, 12]. This possibility may explain the high incidence of AARS after otolaryngological surgery, and patients with upper respiratory tract infection may be at high risk regardless of the presence or absence of neck disease.

AARS has been reported in the postoperative recovery period without evidence of cervical inflammation, including a case following central venous catheter insertion in a child under general anesthesia [13] and cases that developed after surgical procedures in the head [5, 14]. These cases have the common characteristic of rotation of the head under general anesthesia, similar to the three patients in this report. The normal range of cervical rotation is approximately 90° to either side, and more than half of this rotation occurs at the atlantoaxial joint [1, 15]. The physiological range of rotation of the atlas on the axis is 25°–53° to either side [15]. A biomechanical study using CT images from 21 normal children classified rotational behavior of C1 and C2 into three motion phases: (1) C1

moves alone from 0° to 23° with C2 remaining stationary; (2) C1 and C2 move together from 24° to 65°, but C1 always moves at a faster rate, with C2 being pulled by yoking ligaments; and (3) C1 and C2 move in exact union from 65° onward. Head turning thereafter is carried out by subaxial motion only [1]. In another study, the C1–C2 separation angles were within the normal range in 21 cases of AARS, underscoring the argument that it is not an abnormal angle, but rather abnormal dynamics, that characterizes this condition [16]. The pathological stickiness that underlies the disordered dynamics in AARS is a resistance against C1 to close the separation angle and ultimately to recross C2; this abnormal stickiness is present only on counter-rotation [16]. The lower lip of the C1 lateral mass, a piece of detached cartilage, a sprained, infolding synovium, or a lax joint capsule may prevent reversal of the C1–C2 turn cycle [16]. Torticollis may be masked by compensated counter-rotation of the lower cervical spine, with compensatory occiput–C1 laxity in chronic AARS [7]. Neurological compromise is unusual in an atraumatic presentation, unless there is associated anterior or posterior atlantoaxial displacement [7, 13, 15].

Sternomastoid muscle spasm occurs on the side of the chin in AARS, but on the contralateral side in muscular torticollis, and can therefore be used as a clinical feature for differentiating between these conditions [2, 7, 13, 15, 17]. However, in case 2 in this article, sternomastoid muscle spasm occurred on the contralateral side, but AARS was present and posterior arthrodesis of C1–C2 was finally required. This patient was diagnosed with muscular torticollis after the first operation, and torticollis appeared again after the second operation. A bony and muscular pathogenesis may have been present, and this case suggests that it is difficult to differentiate AARS from muscular torticollis based only on the side of sternomastoid muscle spasm, and that neurosurgical evaluation is necessary.

The high incidence of AARS in children may be related to anatomical differences in childhood and early adolescence, because the status of the peripharyngeal lymphoid tissue is hypertrophic [4] and meniscus-like synovial folds rich in blood vessels are seen in the atlanto-occipital and lateral atlantoaxial joints of children [18], which suggests that inflammation, infolding, or rupture of synovial folds into these joints may be the cause of AARS. Some type of interposing soft tissue within the joint may prevent reverse rotation [7, 15, 19].

General anesthesia and muscle relaxants may contribute to loss of the protective function of the neck musculature and have a destabilizing influence on the C1–C2 joints [5, 7]. Airway manipulation is also a possible cause of AARS. The influence of airway management on the articular capsule or synovial membrane of the cervical spine has not been reported in the literature; therefore, it might be reasonable to exclude airway manipulation as a probable cause of AARS [9]. In our case 1, AARS developed after surgery with an inserted LMA without laryngoscopy using a neuromuscular blocker.

The prognosis of AARS differs with the timing of diagnosis. Early diagnosis leads to improved outcome with nonoperative management [2, 13, 15, 19], whereas treatment delay longer than 2–3 months leads to a more severe form of AARS [7]. AARS with symptoms longer than 3 months before treatment has a nonreducible rate of 50%, with a 50% risk of a requirement for fusion [7]. A powerful deforming snap of the neck may be more prone to render tissues beyond their elastic limits, and the longer these tissues remain overstretched, the less likely they will be to spring back to a normal position [7]. All patients diagnosed as AARS within 3 months after onset should be managed with traction immediately to mitigate the adverse effects of chronicity [7]. Traction will disengage some C1–C2 interlocks and pull infolding tissues out of joints [7]. Some form of cervical immobilization should be used after reduction to help to prevent recurrent subluxation for 3 months [7, 17], because such immobilization allows the stretched or edematous ligaments to heal and return to their normal length [2].

Improved perioperative management may be useful for preventing AARS or allow providing better treatment when AARS does occur. A prospective study of AARS showed that the postsurgical group tended to have more severe forms of fixation than found in Grisel's syndrome [7]. We experienced three cases of postsurgical AARS in the space of a year, including a case that required surgical treatment with posterior C1–C2 fusion. These cases involved extended hospital stays, and no explanation about the risk of AARS as a postoperative complication before surgery became a concern. We could not find any articles about guidelines for prevention and early treatment of a postoperative twisted neck. Therefore, we decided to establish guidelines for prevention and early treatment of postoperative AARS based on our three cases and the literature about AARS. Pediatric surgery, otopharyngeal inflammation, general anesthesia, and extreme rotation of the head are risk factors for development of AARS, and patients with multiple factors are probably at higher risk [3, 7, 20]. Our guidelines require the following four items for high-risk patients: (1) informed consent, (2) preoperative check, (3) appropriate surgical position, and [4] postoperative check. We now use these guidelines for the perioperative care of patients with risk of AARS, as follows.

(1) Informed Consent: Patients at risk for developing AARS should be told that there is always a risk of AARS regardless of precautionary measures, and that early detection leads to improved outcome. The

opposite opinion has been advanced that this should not be part of preoperative informed consent because of the rareness of the complication [21]. However, treatment of AARS requires extension of the hospital stay and surgery in some cases, and these unexpected consequences are a potential source of concern for the patient.

(2) Preoperative Check: The range of cervical rotation should be checked preoperatively before anesthesia. The range of cervical rotation varies among individuals, and rotation of the neck may extend beyond the normal range under the influence of muscle relaxants, especially in children. The range of cervical rotation is measured as the angle of the two lines, the line connecting the bridge of the nose and occipital tuber and the perpendicular to a line connecting the right and left acromions (Fig. 1).

(3) Appropriate Surgical Position: A surgical position should be used that avoids excessive rotation of the neck, such as a semilateral decubitus position with a roll under the shoulder, a lateral fold of the neck in the decubitus position, and rotation on the operating table. The physiological range of rotation of the atlas on the axis is 25°–53° to either side [15]. Therefore, the rotation of the neck should be less than 60° or limited by individual characteristics determined preoperatively. Inadvertent head compression by the surgeon and passive aggressive rotation and extension of the head should be avoided [3, 13]. By using a surgical safety checklist, anesthetists discuss the surgical position with the surgeon and nurse before anesthetic induction in every case, and the position should be checked by the surgical team during the course of operation.

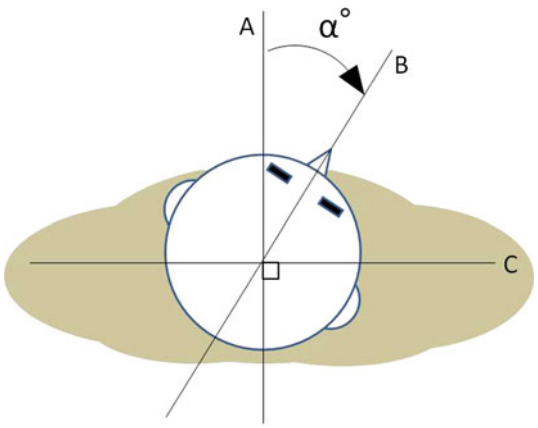

Fig. 1 The range of cervical rotation is the angle of lines *A* and *B*, α°. *A*, perpendicular to a line connecting the right and left acromions; *B*, line connecting the bridge of the nose and occipital tuber; *C*, line connecting the right and left acromions

(4) Postoperative Check: The head position and the range of rotation of the head should be checked at least after recovery from anesthesia and on the day after the operation. Our three cases showed torticollis on the day after surgery, but AARS does not always develop within 24 h [5, 20]. Early recognition of the cervical complication with early neurosurgical consultation is mandatory to prevent serious consequences [6].

We had three cases of postoperative AARS within a short interval of time; therefore, we developed guidelines for prevention and early treatment of postoperative AARS. Postoperative AARS cannot be eliminated completely, but informed consent, preoperative check, appropriate surgical position, and postoperative check may reduce the risk and damage related to this condition.

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