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Perioperative Anaesthetic Management of Pediatric Patients in Craniosynostosis

Kraniosinostozlu Pediatrik Hastalarda Perioperatif Anestezi Yönetimi

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ÖZET

Amaç: Bu çalışmada, hayatı tehdit eden komplikasyonlarla seyredebilen kraniyosinostoz nedeniyle ameliyat edilen çocuk hastalarda perioperatif anestezi yönetimini sunmayı amaçladık.
Gereçler ve Yöntem: Ocak 2009-Ocak 2021 arasında opere edilen 26 kraniyosinostoz hastasının tamamının dosyasını geriye dönük olarak inceledik. Hastaların demografik verileri, ASA skorları, anestezi ve ameliyat süreleri, ek anomali koşulları, hava yolu ve kanama yönetimi ile komplikasyonları analiz edildi.
Bulgular: Çalışmaya alınan 26 hastanın 16'sı trigonosefali, 4'ü skafosefali, 3'ü plajiyosefali ve 3'ü miks tipte idi. Hastaların 20'si (%76.9) erkek, 6'sı (%23.1) kadındı. 26 hastanın beşinde (%19.23) ek anomaliler (1 Apert sendromu, 2 kardiyak anomali ve 2 hidrosefali) vardı. Ortalama ameliyat süresi 167.03 dk ve anestezi süresi 179.92 dk idi. Hastaların direkt laringoskopisinde CL skorları değerlendirildi. Beş hastada (%19.2) CL I, 13 hastada (%50.0) CL II ve 8 hastada (%30.8) CL III bulundu. Ameliyat sırasında 5 hastada (%19.23) şiddetli hipotansiyon gözlendi. Bu hastalara eş zamanlı kan ve sıvı infüzyonu ile 0.03 mg/kg/dk dozunda noradrenalin uygulandı. Ameliyat öncesi ortalama hematokrit değerleri %35.99, ameliyat sırasında %26.85 (0.001) olan hastaların preoperatif ve intraoperatif hematokrit değerleri arasında istatistiksel olarak anlamlı fark bulundu ($p < 0,001$).
Sonuç: Kraniosinostozlu pediatrik hastaların havayolu yönetiminin zor olduğunu ve intraoperatif masif kanama riski olduğunu saptadık. Bu hastalarda dikkatle planlanmış anestezi yönetimi gerekir.

Anahtar Kelimeler: Kraniosinostoz, pediatrik hastalar, anestezi yönetimi, kanama yönetimi

ABSTRACT

Aim: In this study, we aimed to present the perioperative anaesthetic management in pediatric patients who underwent surgery for craniosynostosis which can progress with life-threatening complications.
Materials and Method: We retrospectively reviewed the file of all 26 craniosynostosis patients who were operated between January 2009-January 2021. The following were analyzed retrospectively: demographic data; anesthesia risks; duration of anesthesia and surgery; additional conditions of abnormality; airway and bleeding management; complications.
Results: Of the 26 patients included in the study, 16 had trigonocephaly, 4 scaphocephaly, 3 plagiocephaly, and 3 mixed types. Twenty (76.9%) of the patients were male, and 6 (23.1%) were female. Five out of the 26 patients (19.23%) had additional anomalies (1 Apert syndrome, 2 cardiac anomalies, and 2 hydrocephalus). The mean duration of surgery was 167.03 min, and duration of anesthesia was 179.92 min. CL scores were evaluated in direct laryngoscopy of the patients. CL I was found in 5 patients (19.2%), CL II in 13 patients (50.0%) and CL III in 8 patients (30.8%). Severe hypotension was observed in 5 patients (19.23%) in the intraoperative period. Noradrenaline at a dose of 0.03 mg/kg/min was administered to these patients with simultaneous blood and fluid infusion. A statistically significant difference was found between the preoperative and intraoperative hematocrit values of the patients whose mean preoperative hematocrit values were 35.99% and intraoperatively 26.85% (0.001).
Conclusions: We observed that pediatric patients with craniosynostosis had difficulty in airway management and risk of massive intraoperative bleeding. These patients require carefully planned anesthetic management.

Key words: Craniosynostosis, pediatric patients, anaesthetic management, bleeding management

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INTRODUCTION

Craniosynostosis (CS), which is the early fusion of one or more cranial sutures, is a common craniofacial congenital anomaly requiring surgical treatment (1). CS is seen in approximately 1 in 2000–2500 live births (2). CS occurs in 80% of cases as isolated CS (ICS) and in 20% as part of syndromic CS (SCS). ICS cases are usually simple, that is, not associated with other abnormalities. Their etiology is not genetic; most are probably due to intrauterine fetal head constraint. The growth of the skull is limited, and there is deformity. Surgery should be planned in the early stage, between 3 and 12 months, to prevent the increased intracranial pressure from causing cerebral compression, neurocognitive sequelae, and blindness (1). Hydrocephalus, airway obstruction, small skull compared to the brain or venous drainage anomalies may cause increased intracranial pressure. The required comprehensive surgical operation may result in serious blood loss and transfusion-linked morbidity and may lead to mortality (3). Recently, there has been renewed interest in craniectomy procedures because of the development of minimally invasive endoscopic procedures characterized by lower blood loss, shorter operative times, and reduced hospital stays (4).

CS cases are specific for anesthesia management. Concomitant anomalies, difficulties in airway management, blood product transfusion, and perioperative complications may occur frequently in this patient group (5). SCS is associated with other congenital anomalies involving multiple organ systems. If possible, a comprehensive preoperative evaluation by an otolaryngologist, ophthalmologist, and neuroanesthesiologist is required (3). The airway should be comprehensively evaluated to ensure that the anesthesia technique is well planned in these surgeries. Imaging methods and previous anesthesia history serve as guides in good airway management and intravenous access. The method of anesthesia induction will depend on the experience of the anesthesiologist and, where applicable, the patient and parent preference (1). Considerations in this patient group include the possibility that intravenous access may be difficult, especially in syndromic children (such as excess fatty tissue on the back of the hand); therefore, induction is initiated with inhalation anesthesia, and intravenous access is attempted (1).

Although surgery is specific to the type of synostosis, there are some general principles that apply. These are to prevent progression of the abnormality, correct the abnormality, and reduce the risk of high pressure in cases that have not yet been operated (1).

In this study, we aimed to retrospectively evaluate the anaesthetic management of pediatric patients who underwent CS surgery in a single center for 12 years.

MATERIALS AND METHODS

Data collection: Records of Neurosurgery and Anesthesiology Departments were reviewed, and data from patients who had CS surgery between 2009 and 01.01.2021 were retrospectively analyzed. Data about the patient's demographics, ASA scores, airway and anesthesia management, blood transfusion, durations of surgery and anesthesia, additional abnormalies and complications were recorded.

Anaesthetic management: As a preoperative preparation for difficult endotracheal intubation, some tools were available in the operating room: tubes of all types and sizes; laryngoscope and various blades; Magill forceps; drugs and devices for cardio-pulmonary resuscitation; In addition to percutaneous and surgical tracheostomy sets; fiberoptic bronchoscope; video-laryngoscope; LMA. Standard anesthesia monitoring was performed for all patients admitted to the operating room (ECG, noninvasive and invasive arterial blood pressure, peripheral oxygen saturation, end-tidal CO2, temperature).

The patients were ventilated with 100% O2 for 3 min. After preoxygenation, 2.5 mg/kg propofol and 1 mcg/kg fentanyl were administered intravenously for anesthesia induction. Intravenous 0.6 mg/kg rocuronium bromide was used as a muscle relaxant. After intubation, 2% sevoflurane, 50% O2–air, and 0.25–0.50 mcg/kg/min remifentanil infusion were administered for controlled hypotensive anesthesia.

Arterial pressures were manipulated so that mean arterial pressures were 50-60 mmHg. A vasopressor drug was administered at mean arterial pressures less than 50 mmHg. Isotonic sodium chloride was chosen because there were surgeries in which significant blood loss occurred. Active warming of the patients was provided with a heated blanket. Paracetamol infusion was administered at a dose of 10–15 mg/kg for postoperative analgesia.

The Cormack–Lehane scores, number of endotracheal tubes used, hemodynamic data, and laboratory values of the patients were recorded. The fluid and blood products given to the patients and their amounts were calculated. The following formula was used to calculate the volume of blood to be transfused: estimated blood volume X (ideal hematocrit (HCT) – existing HCT)/HCT of the blood to be transfused.

Surgical procedure: Surgery varies between different centers and according to the type of synostosis. These are extended strip craniectomies, spring-assisted cranioplasty, total calvarial remodeling, frontal orbital advancement and remodeling (FOAR), posterior expansion and remodeling, midface advancement (Le Fort III and monobloc procedures). Surgical techniques suitable for the type of synostosis were selected in our center.

Anesthesia management of all patients was provided by the same experienced neuroanesthesiologists. Surgical

operations were also performed by the same surgical team.

Statistical analysis: The Software Statistical Package for Social Sciences (SPSS) version 18 (SPSS Inc., Chicago, IL, USA) was used for the statistical analysis. Descriptive statistics were used to evaluate the data (mean ± SD). Student's t-test or analysis of variance (ANOVA) was used for the comparison of parametric variables. For the nonparametric variables analysis was performed using chi-square or the Mann–Whitney U test. Statistical significance was established with $p < 0.05$.

RESULTS

Twenty six patients were included in the study. The demographic characteristics of the cases are shown in Table I. Twenty (76.9%) of the 26 cases examined were male and 6 (23.1%) were female. The mean age of the patients was 11 months (1-36 months) and the mean weight was 8.21 kg (3.20-13.50 kg). Five (19.23%) of 26 patients had additional anomalies (1 Apert syndrome, 2 cardiac anomalies and 2 hydrocephalus). Preoperatively, 2 of the patients included in the study were evaluated ASA I (7.7%), 19 ASA II (73.1%),

Figure 2. Trigonocephaly type
A: Preoperative image, B-C: Intraoperative image, D: Postoperative early period image,
E: Postoperative late period image

Table 1. Demographic data

	Mean (Min-Max)		Number
Age (month)	11 (1-36)	Gender	
Weight (kg)	8.21 (3.20-13.50)	Male	20 (76.9%)
		Female	6 (23.1%)
Duration of anesthesia (min)	179.92 (150-200)	ASA	
		I	2 (7.7%)
		II	19 (73.1%)
		III	5 (19.2%)
Duration of Surgery (min)	167.03 (143-190)	Cormach-Lahane Score	
		1	5 (19.2%
		2	13 (50%)
		3	8 (30.8%)
		4	0
Fluid infusion (mL)	210 (70-450)	Central venous catheter	11 (%42.30)

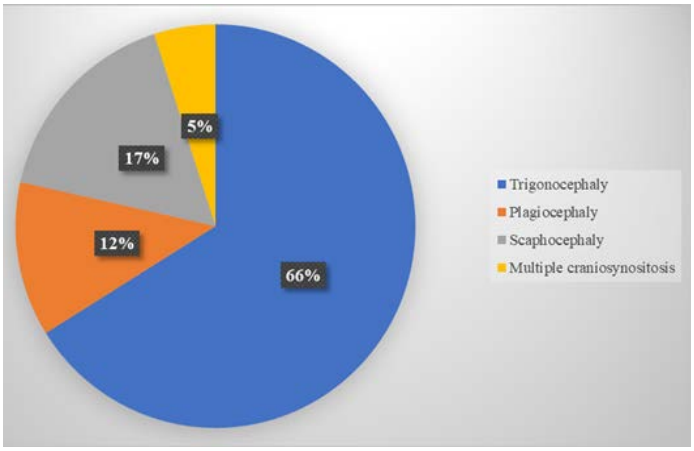

Figure 1. The classification of craniosynositosis of the patients

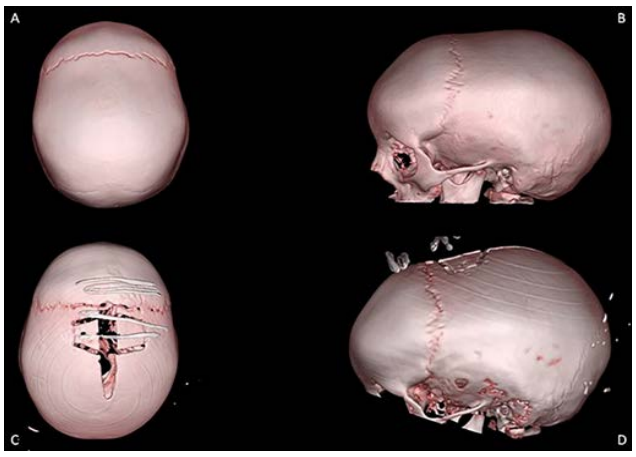

Figure 3. Scaphocephaly type
A-B: Preoperative 3D computed tomography, C-D: Postoperative 3D computed tomography

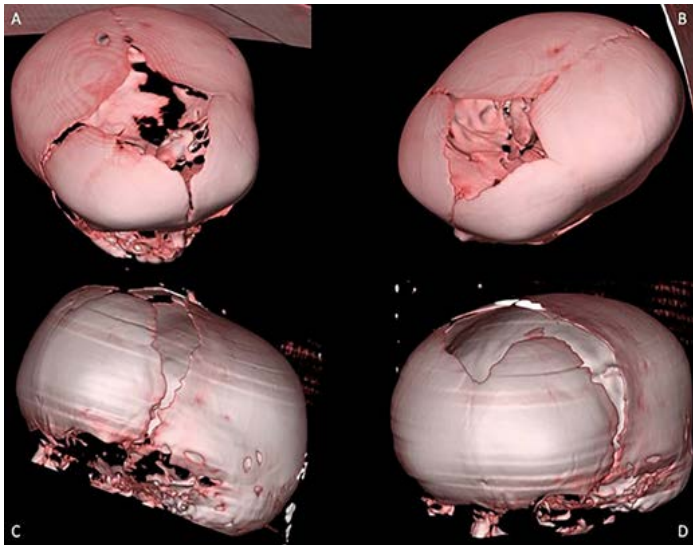

Figure 4. Plagiocephaly (coronal) type.
A-B: Preoperative 3D computed tomography, C-D: Postoperative 3D computed tomography

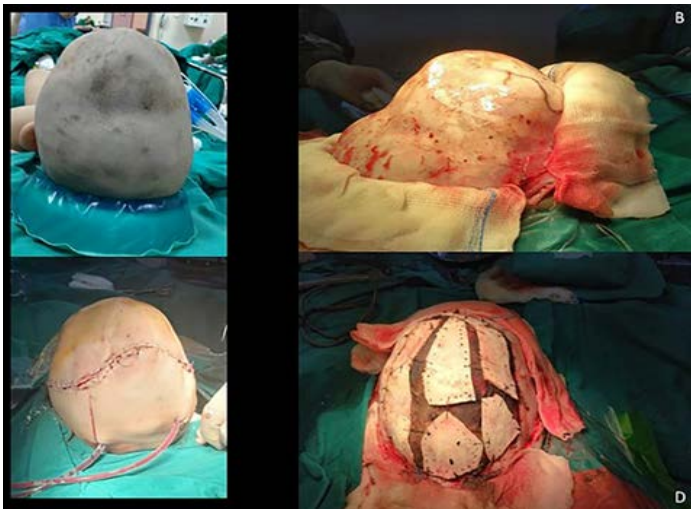

Figure 3. Mix (coronal + sagittal) type.
A: Preoperative image, B: Intraoperative image, C: Postoperative early period image, D: Intraoperative image

and 5 ASA III (19.2%). Mean surgery time was 167.03 minutes, and anesthesia time was 179.92 minutes. Cormack-Lehane (CL) scores were evaluated in direct laryngoscopy of the patients. CL I was found in 5 patients (19.2%), CL II in 13 patients (50.0%) and CL III in 8 patients (30.8%). Eight patients with a score of CL III and evaluated as difficult endotracheal intubation were intubated with a video laryngoscope. No.4 uncuffed endotracheal tube was used in most patients.

Of the 26 patients included in the study, 16 had trigonocephaly, 4 scaphocephaly, 3 plagiocephaly, and 3 mixed types (Figure 1-5). Propofol was used in 69.24% of patients and sevoflurane in 30.76% of patients for anesthesia induction. Sevoflurane induction was preferred in infants younger than 6 months; Propofol induction was preferred in older babies. Severe hypotension was observed in 5 patients (19.23%) in the intraoperative period. Noradrenaline at a dose of 0.03 mg/kg/min was administered to these patients with simultaneous blood and fluid infusion. Preoperative mean hematocrit values of the patients were 35.99%, intraoperative 26.85% (0.001). A statistically significant difference was found between the preoperative and intraoperative hematocrit values of the patients. All intraoperative patients received an average of 20.2 ml/kg of erythrocyte suspension (ES); and 12 patients received 10.8 ml/kg of fresh frozen plasma. Central venous catheterization were performed in 11 patients (42.30%) whose could not inserted adequate vascular access.

DISCUSSION

CS is a condition observed in the first years of life that requires surgical treatment. Bleeding control and difficult airway management are the main difficulties in anesthesia in these patients (5). Difficult endotracheal intubation or failed ventilation is one of the most feared situations for anesthesiologists. Difficult endotracheal intubation is defined as the use of more than one intervention and more than one aid device. Many tests and methods have been developed on this subject and research is still ongoing. The tests and methods used for the pediatric patient group are limited especially for those in the neonatal period. Congenital anomalies found in patients are a strong clue for difficult intubation. Studies show that patients under one year of age, low birth weight, high ASA score, and those who have undergone pediatric cardiac surgery, oral or maxillofacial surgery, have a very high risk of difficult laryngoscopy (6).

In our study, 19.2% of the patients had additional anomalies. One patient had Apert syndrome, two had cardiac anomalies and two had hydrocephalus. Twenty-one of the patients included in the study were one year old or younger. Considering that intubation difficulties will be experienced in patients who have undergone CS surgery, preoperative preparation will facilitate endotracheal intubation and also reduce the complications that may occur. As preoperative preparation for difficult endotracheal intubation: some tools should be available: tubes of all types and sizes; laryngoscope and various blades; Magill forceps; drugs and devices for cardio-pulmonary resuscitation; Besides percutaneous and surgical tracheostomy sets; fiberoptic bronchoscope; video-laryngoscope; LMA. In our study, 8 patients with a CL score of 3 were intubated with a video laryngoscope available in our

clinic.

Video-laryngoscopes are designed for patients whose intubation is expected to be difficult. Considering that attaching an optical system to the tip of the blades provides an indirect view, a different approach can be accepted for intubation and laryngoscopy. These studies have shown that these devices improve the CL laryngoscopic view and can be used as saviors for endotracheal intubation (7,8). The age of the patient, the type of CS, surgery time, and the practices aimed at reducing blood loss during the operation affect the amount of preoperative blood loss. In single suture CS, intraoperative blood loss may be tolerated. However, synostosis and craniofacial interventions involving more than one suture are very risky operations due to complications due to severe bleeding and massive transfusions (1,9). Complications associated with perioperative bleeding include severe hypotension, metabolic acidosis, transfusion reactions, venous air embolism, blood clotting disorders, infections, acute lung injury, cardiac arrest (10). In these patients, blood loss should be closely monitored during the perioperative period. Even in small amounts, bleeding causes severe hypotension and cardiac arrest, especially in newborns. Because this blood constitutes a large part of the blood of newborns. In the first 30 minutes of the surgical procedure, 20-50% of the estimated blood volume is lost (10). Although blood loss during the operation is usually slow, there is constant loss of the venous system and bones.

In our study, preparations were made for blood and anesthesia management by discussing the number of sutures to be corrected before surgery and the expected amount of bleeding. Many different methods have been tried in craniosynostosis surgeries to reduce blood loss and transfusion complications. These; controlled hypotension; minimally invasive and endoscopic procedures; autologous blood donation; use of chemotherapeutic drugs (aminocaproic acid, tranexamic acid) that protect hemostasis; pre-operative recombinant human erythropoietin and fibrin glue (11). However, there is still no accepted routine practice. Controlled hypotension is one of the techniques commonly used in craniofacial surgery. Controlled hypotension improves surgical conditions due to highly vascularized tissues and prevents the operation area from filling with blood. In pediatric patients, a moderate controlled hypotension technique, that is, keeping the mean arterial blood pressure (MAP) at a level of 50-65 mmHg (or a 30% reduction in basal MAP) reduces blood loss and the associated transfusion requirement (12). Studies suggest that this method should be used for blood transfusion during controlled hypotension, due to concerns about cerebral hypoperfusion and decreased oxygen absorption, especially in the presence of anemia. Especially, it is emphasized that even in moderate controlled hypotension, cerebral oxygen uptake may be insufficient and therefore higher FIO2 should be administered. Nicardipine, esmolol, labetalol, sodium nitroprusside, nitroglycerin and remifentanil are agents used for controlled hypotension (13). Studies on the controlled hypotension procedure are ongoing. Short-acting drugs are preferred because they have minimal reubound effects and allow rapid recovery of normal blood pressure after administration (14). In the studies of Shirgoska et al. (15) using sevoflurane and remifentanil to achieve controlled hypotension in children undergoing middle ear surgery, they proved that blood flow in the middle ears of children was reduced and provided good conditions for surgery without the need for additional hypotensive agents. In our study, 0.25-0.50 mcg/kg/min remifentanil infusion was administered to patients for controlled hypotension. Five patients who developed severe intraoperative hypotension were treated with noradrenaline infusion with blood and fluid. Due to controlled hypotension, no serious complications were seen in any of the patients. The superior properties of remifentanil are rapid onset and rapid elimination of its effect, ease of titration, short half-life despite prolonged infusion, and rapid recovery (16,17). Remifentanil infusion is routinely used in patients undergoing both controlled hypotension and total intravenous anesthesia in our clinic.

In our study, a statistically significant difference was observed when preoperative and intraoperative hematocrit values were compared. During the operation, an average of 165.88 ml (30-440 ml) erythrocyte suspension was transfused to the patients. In the study conducted by Küçük et al. (18) the average amount of ES given to patients was 61.2 ml (15-200), while Chocron et al. (19) used an average of 29.6 cc/kg in their study with 100 cases. In the studies of Jeong et al. (2) the mean blood transfusion was determined as 1293 ml. The amount of transfusion depended on the type of surgery. There is also an increased risk to the child because of the smaller blood volume. Surgical correction is recommended for children aged 6 to 12 months, as physiological tolerance to blood loss is better in children older than 6 months (1). The average age of patients operated on in our studies is 11 months. Studies have shown that patients undergoing surgery are usually older than 6 months.

The use of CVP in craniofacial surgeries is not a routine practice in many medical centers. There is no published evidence that monitoring CVP in children is an effective monitoring tool of the intravascular volume status that is useful in preventing hypovolemia (21). Some researchers, who made subjective comments about the value of CVP monitoring data in relation to cases of CS, believe that the use of CVP can help prevent hypotension and hypovolemia (22). In our study, CVP was implanted in 11 patients. In CVP application, patients who were expected to repair more than

one suture and had venous access problems were preferred (to be included in the study). At least two vascular accesses were placed in patients without CVP in order to provide rapid intraoperative blood replacement when necessary.

In our 12-year results in our clinic on patients with CS, no serious complications or patient loss were experienced. Video-laryngoscope was used in difficult airway management. Controlled hypotension was applied to reduce bleeding. CS surgery is a procedure that requires a multidisciplinary approach and well-planned anaesthetic management. Because if complications occur, it can have life-threatening consequences. We believe that the surgery can be successfully performed by using video-laryngoscopy for difficult endotracheal intubation in anesthesia induction, selecting patients of appropriate age and weight, and choosing a controlled hypotension technique for bleeding control.

Etik Kurul: Ethical approval was obtained for this study from the local ethics committee with the decision dated 04.03.2021 and numbered 351.

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