

# POSTOPERATIVNE KOMPLIKACIJE I NJIHOVA KORELACIJA SA OPERATIVNOM TEHNIKOM U LEČENJU GASTROSHIZE

ORIGINALNI RAD

ORIGINAL ARTICLE

## POSTOPERATIVE COMPLICATIONS AND THEIR CORRELATION WITH THE SURGICAL TECHNIQUE IN THE TREATMENT OF GASTROSCHISIS

Natalija Kecman<sup>1</sup>, Vesna Milojković<sup>2</sup>

<sup>1</sup> Univerzitetski klinički centar Srbije, Klinika za hematologiju, Beograd, Srbija

<sup>2</sup> Institut za zdravstvenu zaštitu majke i deteta Srbije „Dr Vukan Čupić”, Beograd, Srbija

<sup>1</sup> University Clinical Center of Serbia, Clinic for Hematology, Belgrade, Serbia

<sup>2</sup> Mother and Child Health Institute of Serbia Dr Vukan Čupić, Belgrade, Serbia

### SAŽETAK

**Uvod:** Gastroshiza predstavlja kongenitalni paraumbilikalni defekt prednjeg trbušnog zida sa evisceracijom abdominalnih organa. Savremeni pristup lečenju gastroshize doprinoje boljim rezultatima, što dokazuje stopa mortaliteta koja iznosi 5% – 10%.

**Cilj rada:** Cilj rada je da se proceni učestalost komplikacija i smrtnog ishoda u populaciji usled gastroshize, kao i povezanost primenjenih hirurških tehnika u lečenju sa istim.

**Materijal i metode:** Retrospektivnom kohortnom studijom obuhvaćeno je 75 bolesnika sa dijagnozom gastroshize, koji su u periodu od 2000. do 2020. godine lečeni u Institutu za zdravstvenu zaštitu majke i deteta Srbije „Dr Vukan Čupić“. Primenom ekskluzivnih kriterijuma dobijen je uzorak sačinjen od 61 bolesnika. Formirane su dve kohorte ispitanika na osnovu primenjene hirurške metode, i to: primarno reponiranje i fascijalno zatvaranje defekta (hirurška kohorta gastroshiza – HKG), odnosno primarno odloženo reponiranje defekta primenom silastične vrećice (silastična kohorta gastroshiza – SKG).

**Rezultati:** Hiruršku kohortu gastroshiza je činilo 38 pacijenata, dok je silastičnu kohortu gastroshiza činilo 23 ispitanika. Utvrđeno je da je nekrotični enterokolitis (engl. *necrotizing enterocolitis* – NEC) bio statistički značajno češća komplikacija kod ispitanika iz silastične kohorte gastroshiza (5/23, odnosno 21,7% u SKG-u i 0/38 u HKG-u; RR 0,32, 95% CI: 0,22 – 0,47;  $p = 0,003$ ). Nije dokazana statistička značajnost razlike u odnosu na učestalost drugih komplikacija: ileus (0/23 u SKG-u i 5/38, odnosno 13,2% u HKG-u; RR 0,59, 95% CI: 0,47 – 0,73,  $p = 0,069$ ), kompartmen sindrom (0/23 u SKG-u i 2/38, odnosno 5,3% u HKG-u; RR 0,61, 95% CI: 0,50 – 0,75,  $p = 0,263$ ) i smrtni ishod (2/23, odnosno 8,7% u SKG-u i 2/38, odnosno 5,3% u HKG-u; RR 1,26, 95% CI: 0,46 – 3,43,  $p = 0,600$ ).

**Zaključak:** Nije utvrđen jedinstveni dokaz o superiornosti jedne metode nad drugom. Rizici od nastanka ileusa i kompartmen sindroma su veći pri primeni tehnike primarnog fascijalnog zatvaranja defekta, dok je rizik od nastanka NEC-a i smrtnog ishoda veći prilikom primene *silo* metode. Izbor metode u lečenju gastroshize zavisi od abdominovisceralne disproporcije, kao i izgleda evisceriranih creva procenjenih pomoću Prognostičkog skora gastroshize (engl. *Gastroschisis Prognostic Score* – GPS).

**Ključne reči:** gastroshiza, komplikacije, primarno fascijalno zatvaranje, primarno odloženo zatvaranje, ishod lečenja

Autor za korespondenciju:

Natalija Kecman

Klinika za hematologiju, Univerzitetski klinički centar Srbije

Dr Koste Todorovića 2, 11000 Beograd, Srbija

Elektronska adresa: kecman.natalija@gmail.com

### ABSTRACT

**Introduction:** Gastroschisis is a congenital paraumbilical defect of the anterior abdominal wall with evisceration of the abdominal organs. A modern approach to treating gastroschisis has contributed to better results, as evidenced by the 5% – 10% mortality rate.

**Aim:** The study aims to evaluate the frequency of complications and death in the population due to gastroschisis, as well as the connection between the surgical techniques used in the treatment and the outcome of the treatment.

**Material and methods:** The retrospective cohort study included 75 patients diagnosed with gastroschisis, treated from 2000 to 2020 at the Mother and Child Health Institute of Serbia Dr Vukan Čupić. By applying the exclusion criteria, a sample of 61 patients was obtained. Two cohorts of subjects were formed based on the applied surgical method, namely: primary repositioning and fascial closure of the defect (gastroschisis surgical cohort – GSC), i.e. delayed primary repositioning of the defect using a silastic bag (silastic gastroschisis cohort – SGC).

**Results:** The gastroschisis surgical cohort comprised 38 patients, while the silastic gastroschisis cohort comprised 23 subjects. It was found that necrotizing enterocolitis (NEC) was a statistically significantly more frequent complication in subjects from the silastic gastroschisis cohort (5/23, i.e. 21.7% in SGC and 0/38 in GSC; RR 0.32, 95% CI: 0.22 – 0.47;  $p = 0.003$ ). The statistical significance of the difference in relation to the frequency of other complications was not proven: ileus (0/23 in SGC and 5/38, i.e. 13.2% in GSC; RR 0.59, 95% CI: 0.47 – 0.73,  $p = 0.069$ ), compartment syndrome (0/23 in SGC and 2/38, i.e. 5.3% in GSC; RR 0.61, 95% CI: 0.50 – 0.75,  $p = 0.263$ ) and death (2/23, i.e. 8.7% in SGC and 2/38, i.e. 5.3% in GSC; RR 1.26, 95% CI: 0.46 – 3.43,  $p = 0.600$ ).

**Conclusion:** There is no distinctive proof of the superiority of one method over another. The risks of ileus and compartment syndrome are higher when applying primary fascial closure of the defect, while the risks of NEC and fatal outcome are higher when the *silo* method (use of a silastic bag, i.e., silo bag) is applied. The choice of method in treating gastroschisis depends on the abdominovisceral disproportion and the physical appearance of the eviscerated intestines, assessed with the Gastroschisis Prognostic Score (GPS).

**Keywords:** gastroschisis, complications, primary fascial closure, delayed primary closure, treatment outcome

Corresponding author:

Natalija Kecman

Clinic for Hematology, University Clinical Center of Serbia

2 Dr Koste Todorovića Street, 11000 Belgrade, Serbia

E-mail: kecman.natalija@gmail.com

Primljeno • Received: March 6, 2024;

Revidirano • Revised: May 10, 2024;

Prihvaćeno • Accepted: May 17, 2024;

Online first: June 25, 2024

DOI: 10.5937/smcl5-49697

## UVOD

Gastroshiza predstavlja kongenitalni paraumbilikalni defekt prednjeg trbušnog zida sa evisceracijom abdominalnih organa. Prisutna je kod 1 – 4 : 10.000 novorođenčadi [1]. Češće se javlja kod muškog pola i prematu-rusa. Za razliku od ostalih defekata prednjeg trbušnog zida, gastroshiza obično nije udružena sa hromozom-skim ili drugim strukturnim anomalijama, osim atrezije ili stenoze creva, koje se viđaju u oko 10% slučajeva. Najčešće su eviscerirane vijuge tankog i debelog creva, ali mogu biti prisutni i deo želuca, ovarijumi, tube, te-stisi, deo mokraćne bešike. Kao posledica evisceracije organa dolazi do povećanog gubitka tečnosti i sklo-nosti novorođenčeta ka hipotermiji. Creva su sjajna, „lakirana“, zadebljala, skraćena, pokrivena eksudatom, međusobno adherirana. Znaci iritativnog peritoniti-sa se javljaju zbog izlaganja creva dejstvu amnionske tečnosti. Jedan od primarnih problema gastroshize je relativno nerazvijena abdominalna duplja u koju se ne mogu smestiti eviscerirani organi bez značajnijeg povećanja intraabdominalnog pritiska. Sklonost paraumbilikalnog defekta da se skupi značajno doprinosi poremećaju vaskularizacije evisceriranih creva [2,3].

Savremeni pristup u hirurškom lečenju gastroshize doprineo je boljim rezultatima, što se može videti iz sto-pe mortaliteta koja ne prelazi 5% – 10% [4]. Komplikovane gastroshize (udružene sa atrezijom, volvulusom, ishemijom, stenozom, nekrozom, perforacijom creva) imaju loš ishod, sa stopom mortaliteta 19% – 30%. Kla-sičan stav i pristup u lečenju gastroshize podrazumeva primarno manuelno reponiranje evisceriranih organa i suturiranje defekta u dva sloja (fascija i koža), u opštoj anesteziji. Primarno odloženo, odnosno etapno reponiranje je neophodno kada hirurg ne može da reponira organe bez značajnijeg povećanja intraabdominalnog pritiska. Tada se primenjuje silastična vreća sa ojačanim prstenom, koje omogućava gravitaciono reponiranje creva [5–7].

Najznačajnija životno ugrožavajuća komplikacija primarnog manuelnog reponiranja evisceriranih organa i fascijalnog suturiranja defekta je razvoj abdominalnog kompartiment sindroma, koji je definisan kao produženo povišenje intraperitonealnog pritiska iznad 20 mmHg (normalno je oko 6,5 mmHg), uz razvoj multiorganskih disfunkcija [8–10]. U faktore rizika spadaju: smanjena komplijansa abdominalnog zida, povećana crevna intraluminalna zapremina, povećana abdominalna zapremina i povećana kapilarna permeabilnost [11]. Abdominalni kompartiment sindrom je definisan prisustvom sledećih kliničkih znakova: povećana si-stemska i plućna vaskularna rezistencija, oligurija, hipoksija, smanjen minutni volumen, hipotenzija, acidoz-a [12].

## INTRODUCTION

Gastroschisis is a congenital paraumbilical defect of the anterior abdominal wall with evisceration of abdominal organs. It occurs in 1 – 4 : 10,000 newborns [1]. It happens more often in males and premature infants. Unlike other anterior abdominal wall defects, gastroschisis is not usually associated with chromosomal or other structural anomalies, except for intestinal atresia or stenosis, which are seen in about 10% of cases. Usually, small and large intestine loops are eviscerated, however, a part of the stomach, ovaries, fallopian tubes, testicles, and urinary bladder can also be involved. Because of the evisceration of the organs, there is an increased loss of fluid and a susceptibility of the newborn to hypothermia. The intestines are glossy in appearance, with thickened walls. They are shortened, covered in exudate, with adhesions between the loops. Signs of irritative peritonitis occur due to the exposure of the intestine to the amniotic fluid. One of the primary problems of gastroschisis is the relatively underdeveloped abdominal cavity, which cannot accommodate the eviscerated organs without a significant increase in intra-abdominal pressure. The propensity of the paraumbilical defect to contract significantly contributes to abnormal vascularization of the eviscerated intestines [2,3].

The contemporary approach to the surgical treatment of gastroschisis has contributed to better results, which is evident from the mortality rate that does not exceed 5% – 10% [4]. Complicated gastroschisis (associated with atresia, volvulus, ischemia, stenosis, necrosis, and intestinal perforation) has a poor outcome, with a mortality rate of 19% – 30%. The traditional approach to the treatment of gastroschisis involves primary manual repositioning of the eviscerated organs and double-layered suturing of the defect (fascia and skin), under general anesthesia. Delayed primary or staged repositioning is necessary when the surgeon cannot reposition organs without a resulting significant increase in intra-abdominal pressure. In such cases, a silo bag with a reinforced loop is applied, enabling gravitational repositioning of the intestines [5–7].

The most significant life-threatening complication of primary manual repositioning of eviscerated organs and fascial suturing of the defect is the development of abdominal compartment syndrome, which is defined as a prolonged increase in intraperitoneal pressure above 20 mmHg (the normal pressure is around 6.5 mmHg), with the development of multiorgan dysfunction [8–10]. Risk factors include the following: reduced compliance of the abdominal wall, increased intestinal intraluminal volume, increased abdominal volume, and increased capillary permeability [11]. Abdominal compartment syndrome is defined by the presence

Abdominalna hipertenzija je uzrok razvoja ishemije, masivne nekroze, perforacije creva, nastanka renalne i respiratorne insuficijencije i sepsa, a sa tim u vezi i uzročno-posledične povezanosti razvoja sindroma sistemskog inflamatornog odgovora (engl. *systemic inflammatory response syndrome – SIRS*) i sindroma multiple organske disfunkcije (engl. *multiple organ dysfunction syndrome – MODS*) [8,13,14]. Osim toga, povišen intraabdominalni pritisak može da dovede i do nastanka gastroezofagealnog refluksa, intolerancije enteralnog unosa, malapsorpcije i pseudoopstrukcije. Intolerancija i malapsorpcija su česti zbog patofiziološke prirode postojanja relativno kratkog creva, dok se razvoj pseudoopstrukcije, koja može progredirati do ileusa, manifestuje distenzijom trbuha, dilatacijom crevnih vijuga sa stazom crevnog sadržaja, porastom broja bakterija i razvojem infekcije, pa i sepsu [15].

Još jedan od značajnih faktora morbiditeta i mortaliteta u populaciji sa gastoschizom je nastanak nekrotičnog eneterokolitisa (engl. *necrotizing enterocolitis – NEC*). Literatura pokazuje da je klasičan *NEC*, pa i onaj udružen sa gastoschizom, upravo posledica klasičnih karakteristika bolesnika – prematurusi niske gestacione starosti, male porođajne telesne mase [16], sa znacima asfiksije i hipovolemije, nastankom transmuralnih ishemičnih promena u crevima, a potom i nastankom direktnе translokacije bakterija u limfotok i cirkulaciju, što može dovesti do pojačanog *SIRS-a*, kao i do *MODS-a* [17,18]. Noviji literaturni podaci za poslednjih pet godina ukazuju da je ukupna incidencija *NEC-a* smanjena u poređenju sa ranijim izveštajima, sa 15% – 20% [19] na današnjih 3% – 5% [16,20,21]. *NEC* kod gastoschize nema svoje klasične patofiziološke mehanizme, razlikuje se u poređenju sa tradicionalnim *NEC-om*, jer je odložen, kasniji i češće je posledica postoperativnog kompartment sindroma, odnosno kompromitovane crevne cirkulacije i ishemije, ali i odložene postoperativne inicijacije enteralne ishrane, ili prolongiranog gladovanja udruženog sa prekomernim rastom bakterija i razvojem infekcije, koja je posebno ugrožavajuća ukoliko je klostridijalnog porekla. Smatra se da blagovremeni enteralni unos majčinog mleka deluje protektivno i smanjuje rizik od *NEC-a* posle operativnog zbrinjavanja gastoschize [21-23].

Sa druge strane, eksteriorizovana creva, sa udruženom malrotacijom i *mesenterium commune*, sklona su torziji i volvulusu srednjeg creva, što može dovesti do poremećaja mezenterijalne cirkulacije, smanjenja protoka krvi i gangrene zahvaćenog creva. Nekada je dovoljna samo prekomerna zategnutost krvnih sudova mezenterijuma, ili kompresija od strane ivica defekta na prednjem trbušnom zidu, ili tesna silastična vreća, pa da dođe do kompromitacije mezenterijalne cirku-

of the following clinical signs: increased systemic and pulmonary vascular resistance, oliguria, hypoxia, decreased cardiac output, hypotension, and acidosis [12].

Abdominal hypertension is the cause of the development of ischemia, massive necrosis, intestinal perforation, renal and respiratory insufficiency, and sepsis, and by extension, it results in the cause-and-effect relationship in the development of systemic inflammatory response syndrome (SIRS) and multiple organ dysfunction syndrome (MODS) [8,13,14]. In addition, elevated intra-abdominal pressure can lead to gastroesophageal reflux, intolerance of enteral intake, malabsorption, and pseudo-obstruction. Intolerance and malabsorption are common due to the pathophysiological nature of the relatively short intestine, while the development of pseudo-obstruction, which can further progress to ileus, manifests as distension of the abdomen, dilation of the intestinal loops with intestinal stasis, an increase in the number of bacteria, and the development of infection, and even sepsis [15].

Another significant factor of morbidity and mortality in the population with gastoschisis is the development of necrotizing enterocolitis (NEC). Literature shows that typical NEC, including NEC associated with gastoschisis, is, in fact, the direct consequence of the typical characteristics of the patient, i.e., preterm infants of low gestational age and low birth weight [16] with signs of asphyxia and hypovolemia, exhibiting transmural ischemic changes in the intestines and consequently direct translocation of bacteria into the lymph flow and blood circulation, which can lead to increased SIRS, as well as to MODS [17,18]. Recent literature data, pertaining to the last five years, indicate that the overall incidence of NEC has decreased, as compared to earlier reports, i.e., from 15% – 20% [19] to 3% – 5%, as it is today [16,20,21]. NEC in gastoschisis does not have its typical pathophysiological mechanisms, it differs from typical NEC because it is delayed, occurs later, and is more often a consequence of postoperative compartment syndrome, i.e., of compromised intestinal circulation and ischemia, but also of delayed postoperative initiation of enteral nutrition, or of prolonged starvation associated with the excessive multiplication of bacteria and the development of an infection, which is especially dangerous if it is of clostridial origin. Timely enteral intake of breast milk is believed to be protective and to reduce the risk of NEC after operative treatment of gastoschisis [21-23].

On the other hand, exteriorized intestines, with associated malrotation and *mesenterium commune*, are prone to torsion and volvulus of the midgut, which can lead to mesenteric circulation disorders, decreased blood flow, and gangrene of the affected intestine.

laciјe. Crevne vijuge mogu zahtevati odvrtanje i adekvatno slaganje u abdomen ili *silo bag* da bi se izbegla strangulacija i gangrena, a ako je abdominalni defekt mali u poređenju sa masom eksteriorizovanih creva, neophodno ga je odmah proširiti da bi se izbegla vaskularna kompromitacija [24].

Lečenje kompleksnih formi gastroshize praćeno je mnogobrojnim komplikacijama: potrebom za ponovljenim laparotomijama, sindromom ultrakratkog creva, dugotraјnom parenteralnom nutricijom, holestaznom insuficijencijom jetre, sepsom i visokim troškovima lečenja [5,25].

Cilj rada je ocena učestalosti komplikacija i smrtnog ishoda u populaciji sa gastroshizom i procena povezanosti primenjenih hirurških tehnika u lečenju sa istim.

## MATERIJALI I METODE

Ispitivanu populaciju pacijenata sa gastroshizom je činilo 75 bolesnika. Na osnovu ekskluzionih kriterijuma (ishemija, stenoza, nekroza, perforacija creva prisutna na rođenju, prisustvo atrezije creva, agenezija tankog creva i kolona, mali defekt prednjeg trbušnog zida u koji ne može da se smesti silastična vreća), načinjena je selekcija, te se studija nije odnosila na 14 (14/75) ispitanika, pa je u studijskom uzorku ostao 61 (61/75) ispitanik.

Istraživanje je urađeno u formi retrospektivne kohortne studije. Uzorak u istraživanju čine novorođenčad sa dijagnozom gastroshize koja su u periodu između 2000. i 2020. godine lečena na Institutu za zdravstvenu zaštitu majke i deteta Srbije „Dr Vukan Čupić“ (IMD). IMD predstavlja referentnu ustanovu za prenatalno dijagnostikovanje i lečenje ove kongenitalne anomalije.

Podaci za analizu su prikupljeni na osnovu medicinske dokumentacije, postojećih trajnih istorija bolesti, koje sadrže sve podatke od prijema bolesnika na Odeljenje hirurške neonatalne intenzivne terapije do vremena otpusta kući ili premeštaja na Odeljenje pedijatrijske intenzivne nege.

Do decembra 2006. godine, primenjivala se hirurška tehnika manuelnog reponiranja evisceriranih organa i primarno fascijalno zatvaranje defekta u opštoj anesteziji. Ovu grupu činilo je 38 bolesnika i oni predstavljaju hiruršku kohortu gastroshiza (HKG). Od prvog januara 2007. godine, primenjuje se i novi pristup – primarno odloženo zatvaranje defekta bez opšte anestezije, odnosno gravitaciono reponiranje primenom silastične vreće sa ojačanim prstenom. Ovu grupu činilo je 23 bolesnika i oni predstavljaju silastičnu kohortu gastroshiza (SKG). Varijable koje se odnose na karakteristike gastroshize i na izbor hirurške tehnike su određene tipom komplikacija: kompartmen sindrom, ileus, NEC, smrtni ishod.

Sometimes, the excessive tightness of the blood vessels of the mesenterium, compression by the edges of the defect on the front abdominal wall, or a tight silo bag can be enough to compromise the mesenteric circulation. Intestinal loops may require unwinding and appropriate placing in the abdomen or silo bag to avoid strangulation and gangrene, and if the abdominal defect is small, as compared to the mass of the exteriorized intestines, it is necessary to dilate it immediately, to avoid vascular compromise [24].

The treatment of complex forms of gastroshisis is accompanied by numerous complications: the need for repeated laparotomies, ultra-short bowel syndrome, long-term parenteral nutrition, cholestatic liver failure, sepsis, and high treatment costs [5,25].

The study aims to assess the frequency of complications and fatal outcome in the population with gastroshisis and to assess the association between the surgical techniques applied in the treatment of gastroshisis and the occurrence of the said complications, i.e., fatal outcome.

## MATERIALS AND METHODS

The studied population of patients with gastroshisis consisted of 75 patients. Based on the exclusion criteria (ischemia, stenosis, necrosis, perforation of the intestine present at birth, presence of intestinal atresia, agenesis of the small intestine and colon, a small defect of the anterior abdominal wall wherein the silo bag cannot be placed), a selection was made, wherefore the study did not apply to 14 (14/75) respondents. Thereby, 61 (61/75) respondents remained in the study sample.

This is a retrospective cohort study. The study sample consists of newborns diagnosed with gastroshisis, treated between 2000 and 2020, at the Mother and Child Health Institute of Serbia Dr Vukan Čupić. This is the reference hospital for prenatal diagnosis and treatment of this congenital anomaly.

Data for analysis were collected from medical records, existing permanent medical histories that contain all the data collected from patient admission to the Department of Surgical Neonatal Intensive Care until the time of discharge or transfer to the Department of Pediatric Intensive Care.

Until December 2006, the surgical technique of manual repositioning of eviscerated organs and primary fascial closure of the defect under general anesthesia was applied. The group of patients treated in this way consisted of 38 patients comprising the gastroshisis surgical cohort (GSC). As of January 1, 2007, a new approach has been applied – delayed primary closure of the defect without general anesthesia, i.e. gravitational repositioning using a silo bag with a re-

Prikupljeni podaci su uneti u kompjutersku bazu podataka, a za obradu podataka je korišćen program SPSS 2016. Ocena učestalosti komplikacija i smrtnog ishoda data je u vidu 95%-og intervala poverenja (engl. 95% confidence interval – CI). Značajnost razlike je testirana parametarskim Student t-testom (kod pravilne distribucije podataka) i Men-Vitnijevim U testom (kod nepravilne distribucije podataka). Podaci su smatrani statistički značajnim za  $p < 0,05$ . Za procenu povezanosti hirurških tehnika sa ishodom komplikacija upotrebljen je atributivni rizik (AR) i relativni rizik (RR). Rezultati su predstavljeni tekstualno, tabelarno i grafički.

## REZULTATI

Podaci koji se odnose na učestalost i stope komplikacija su prikazani u **Tabeli 1**. NEC je registrovan kod 5 od 23 (21,7%) pacijenata, kod kojih je primenjena tehnika zatvaranja silastičnom vrećicom, dok kod hirurškog zatvaranja nije registrovan nijedan slučaj (0/38). Ileus se razvio kod 5 od 38 (13,2%) pacijenata tretiranih hirurškom tehnikom, dok ove komplikacije u drugoj kohorti nije bilo (0/23 u SKG-u). Kompartiment sindrom je razvilo dva od 38 (5,3%) pacijenata tretiranih primarnim fascijalnim zatvarnjem, ali nije registrovan nijedan slučaj ove komplikacije u kohorti gde je primenjeno zatvaranje defekta silastičnom vrećicom (0/23 u SKG-u). Smrtni ishod je zabeležen kod dva od 38 (5,3%) pacijenata u hirurškoj kohorti gastroshiza i kod dva od 23 (8,7%) pacijenata u silastičnoj kohorti gastroshiza.

Zaključeno je da nije postojala statistički značajna razlika između primarnog reponiranja i fascijalnog zatvaranja defekta, u odnosu na primenu silastične vreće, a prema učestalosti nastanka ileusa ( $p = 0,069$ ), kompartiment sindroma ( $p = 0,263$ ) i učestalosti smrtnog ishoda ( $p = 0,600$ ). Sa druge strane, utvrđena je statistički značajna razlika između primarnog reponiranja i fascijalnog zatvaranja defekta i primene silastične vreće, a prema učestalosti NEC-a. NEC je bio statistički značajno češća komplikacija kod ispitanika

inforced loop. This group consisted of 23 patients comprising the silastic gastroshisis cohort (SGC). Variables related to the characteristics of gastroshisis and the choice of surgical technique were determined by the type of complication: compartment syndrome, ileus, NEC, and fatal outcome.

The data collected were entered into a computer database, and the SPSS 2016 program was used for data processing. The assessment of the frequency of complications and fatal outcome was given in the form of a 95% confidence interval (CI). The significance of the difference was tested with the parametric Student t-test (for normal data distribution) and the Mann-Whitney U test (for non-normal data distribution). Data were considered statistically significant for  $p < 0.05$ . The attributable risk (AR) and the relative risk (RR) were used to assess the association between surgical techniques and complications, as an outcome. The results are presented textually, tabularly, and graphically.

## RESULTS

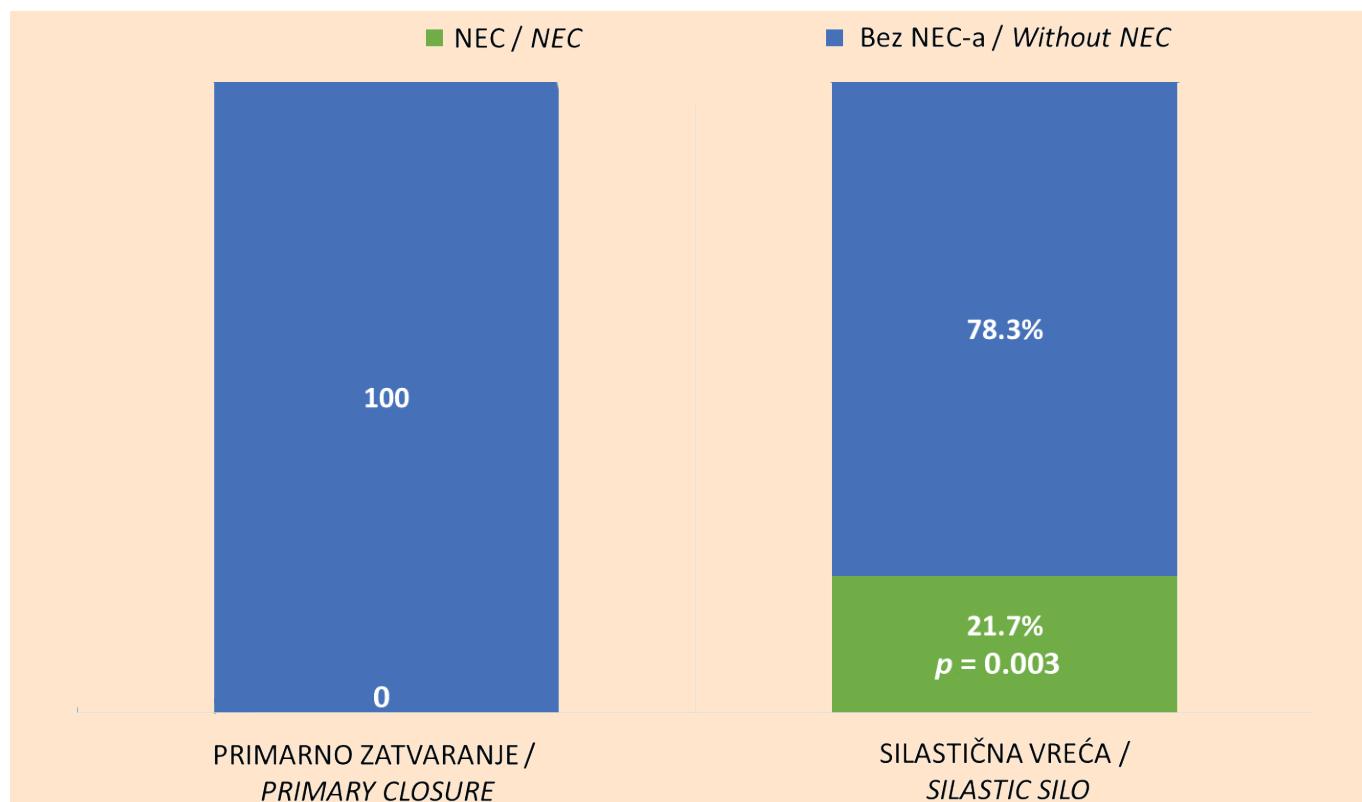
Data related to the frequency and rate of complications are presented in **Table 1**. NEC was registered in 5 out of 23 (21.7%) patients, in whom the silo bag closure technique was applied, while no cases of NEC were registered with surgical closure (0/38). Ileus developed in 5 out of 38 (13.2%) patients treated with the surgical technique, while this complication did not occur in the second cohort (0/23 in SGC). Compartment syndrome developed in two out of 38 (5.3%) patients treated with primary fascial closure, but no cases of this complication were registered in the cohort where closure of the defect with a silo bag was applied (0/23 in SGC). Fatal outcome was registered in two out of 38 (5.3%) patients in the gastroshisis surgical cohort and two out of 23 (8.7%) patients in the silastic gastroshisis cohort.

The conclusion is that there was no statistically significant difference between primary repositioning and fascial closure of the defect, in relation to the application of a silo bag, and in terms of the frequency of

**Tabela 1.** Učestalost komplikacija i smrtnog ishoda u zavisnosti od primenjene hirurške tehnike

**Table 1.** Frequency of complications and fatal outcome, depending on the surgical technique

Komplikacije / Complications	Primarno zatvaranje / Primary closure (n = 38) HKG / GSC	Silastična vreća / Silastic silo (n = 23) SKG / SGC	p
Ileus / Ileus	5 (13.2%)	0	0.069
NEC / NEC	0	5 (21.7%)	0.003
Kompartiment sindrom / Compartment syndrome	2 (5.3%)	0	0.263
Smrtni ishod / Fatal outcome	2 (5.3%)	2 (8.7%)	0.600



**Grafikon 1.** Prikaz učestalosti NEC-a u hirurškoj kohorti gastroshize i silastičnoj kohorti gastroshize

silastične kohorte gastroshiza ( $p = 0,003$ ). Ova statistička značajnost prikazana je u **Grafikonu 1**.

Za procenu povezanosti hirurških tehnika sa komplikacijama i smrtnim ishodom, upotrebljen je atributivni (apsolutni) rizik (AR) i relativni rizik (RR), koji su dati u vidu 95%-og intervala poverenja (95% CI). Ovi podaci su prikazani u **Tabeli 2**. Zaključeno je da je atributivni rizik od ileusa koji je posledica izloženosti određenoj hirurškoj tehnici, u HKG-u iznosio 13%, a u SKG-u 0%, sa 95% CI. Atributivni rizik od nastanka ileusa je bio 13% u studijskom uzorku i bio je povezan je sa tehnikom primarnog fascijalnog zatvaranja, sa 95% CI. U studijskom uzorku, relativni rizik od nastanka ileusa

**Tabela 2.** Apsolutni rizik (AR) i relativni rizik (RR) od komplikacija i smrtnog ishoda

Komplikacije / Complications	Hirurška tehnika / Surgical technique		RR (95% CI)
	AR (95% CI)	Silastična vreća / Silastic silo	
Primarno zatvaranje / Primary closure			
Ileus / Ileus	0.13 (0.04 - 0.28)*	0 (0 - 0)	0.59 (0.47 - 0.73)*
NEC / NEC	0 (0 - 0)	0.22 (0.07 - 0.44)	0.32 (0.22 - 0.47)
Kompartment sindrom / Compartment syndrome	0.05 (0.01 - 0.18)*	0 (0 - 0)	0.61 (0.50 - 0.75)*
Smrtni ishod / Fatal outcome	0.05 (0.01 - 0.18)*	0.09 (0.01 - 0.28)	0.74 (0.26 - 2.08)* 1.26 (0.46 - 3.43)*

\* Vrednosti u zagradama označenim \* odnose se na intervale vrednosti u populaciji

\* Values in parentheses marked with \* refer to intervals of values in the population

je bio 0,59 puta veći u hirurškoj kohorti nego u silastičnoj kohorti gastroshiza, sa 95% CI. Relativni rizik od nastanka ileusa u populaciji je bio 0,47 – 0,73 puta veći pri primeni tehnike primarnog fascijalnog zatvaranja defekta, sa 95% CI.

Atributivni rizik od NEC-a nastalog usled izloženosti određenoj hirurškoj tehnici, u HKG-u je iznosio 0%, a u SKG-u 22%, sa 95% CI. Atributivni rizik od nastanka NEC-a u studijskom uzorku je iznosio 22% i bio je povezan sa tehnikom primene silastične vreće, sa 95% CI. U populaciji je incidencija nastanka NEC-a, kao komplikacije kod pacijenata sa gastroshizom, bila 4% – 18,5% [22]. Relativni rizik od pojave NEC-a je bio 0,32 puta veći u SKG-u nego u HKG-u, sa 95% CI. U populaciji, relativni rizik od nastanka NEC-a je bio 0,22 – 0,47 puta veći pri primeni tehnike silastične vreće nego pri primeni primarnog manuelnog reponiranja i fascijalnog zatvaranja defekta, sa 95% CI.

Atributivni rizik od pojave kompartment sindroma uzrokovanoj izloženošću određenoj hirurškoj tehnici, u HKG-u je iznosio 5%, a u SKG-u 0%, sa 95% CI. Atributivni rizik od nastanka kompartment sindroma je u studijskom uzorku bio 5%, i bio je posledica primene primarnog reponiranja evisceriranih organa i fascijalnog zatvaranja defekta, sa 95% CI. Relativni rizik od pojave kompartment sindroma je bio 0,61 puta veći u HKG-u, u odnosu na SKG, sa 95% CI. U populaciji, relativni rizik od nastanka kompartment sindroma je bio 0,50 – 0,75 puta veći pri primeni tehnike primarnog reponiranja evisceriranih organa i fascijalnog zatvaranja defekta nego pri primeni tehnike silastične vreće, sa 95% CI.

Atributivni rizik od smrtnog ishoda, nastalog usled izloženosti određenoj hirurškoj tehnici, u HKG-u je iznosio 5%, a u SKG-u 9%, sa 95% CI. U populaciji, absolutni rizik od nastanka smrtnog ishoda usled komplikacije, povezan sa tehnikom primarnog reponiranja evisceriranih organa i fascijalnog zatvaranja defekta, iznosio je 1% – 18%, sa 95% CI, dok je apsolutni rizik povezan sa tehnikom primene silastične vreće iznosio 1% – 28%, sa 95% CI. Relativni rizik od smrtnog ishoda je bio 1,26 puta veći u SKG-u nego u HKG-u, sa 95% CI. U populaciji, relativni rizik nastanka smrtnog ishoda kao komplikacije je bio 0,46 – 3,43 puta veći u SKG-u nego u HKG-u, sa 95% CI. Ipak, u studijskom uzorku nije postojala statistička značajnost razlike silastične kohorte gastroshize u odnosu na hiruršku kohortu gastroshize, a prema učestalosti smrtnog ishoda.

## DISKUSIJA

Za adekvatan tretman gastroshize potrebna je integracija više disciplina (hirurgija, ginekologija i akušerstvo, neonatologija, pedijatrija). Gastroshizu je moguće dijagnostikovati na redovnim ultrazvučnim pregledima

concluded that the attributable risk of ileus resulting from exposure to a certain surgical technique, was 13% in GSC and 0% in SGC, with a 95% CI. The attributable risk of ileus was 13% in the study sample and was associated with the primary fascial closure technique, with a 95% CI. In the study sample, the relative risk of developing ileus was 0.59 times higher in the surgical cohort than in the silastic gastroshisis cohort, with a 95% CI. The relative risk of ileus in the population was 0.47 – 0.73 times higher when the technique of primary fascial closure of the defect was applied, with a 95% CI.

The attributable risk of NEC due to exposure to a particular surgical technique was 0% in GSC and 22% in SGC, with a 95% CI. The attributable risk of NEC in the study sample was 22% and was associated with the silastic bag application technique, with a 95% CI. In the population, the incidence of NEC, as a complication in patients with gastroschisis, was 4% – 18.5% [22]. The relative risk of developing NEC was 0.32 times higher in SGC than in GSC, with a 95% CI. In the population, the relative risk of developing NEC was 0.22 – 0.47 times higher with the application of the silo bag technique than with the application of primary manual repositioning and fascial closure of the defect, with a 95% CI.

The attributable risk of compartment syndrome caused by exposure to a particular surgical technique was 5% in GSC and 0% in SGC, with a 95% CI. The attributable risk of compartment syndrome in the study sample was 5%, and was the consequence of the application of primary repositioning of eviscerated organs and fascial closure of the defect, with a 95% CI. The relative risk of the occurrence of compartment syndrome was 0.61 times higher in GSC, as compared to SGC, with a 95% CI. In the population, the relative risk of compartment syndrome was 0.50 – 0.75 times higher when applying the technique of primary repositioning of eviscerated organs and fascial closure of the defect than when applying the silo bag technique, with a 95% CI.

The attributable risk of fatal outcome resulting from exposure to a particular surgical technique was 5% in GSC, and 9% in SGC, with a 95% CI. In the population, the absolute risk of fatal outcome due to complications, which was associated with the technique of primary repositioning of eviscerated organs and fascial closure of the defect, was 1% – 18%, with a 95% CI, while the absolute risk associated with the silo bag application technique was 1% – 28%, with a 95% CI. The relative risk of fatal outcome was 1.26 times higher in SGC than in GSC, with a 95% CI. In the population, the relative risk of death as a complication was 0.46 – 3.43 times higher in SGC than in GSC, with a 95% CI. However, in the study sample, there was no statistical significance in the difference between the silastic gas-

u toku trudnoće, kao i određivanjem alfa-fetoproteina u serumu majke (skrining), što ukazuje na veliki značaj prenatalne zaštite. U razvijenim zemljama, broj ovako otkrivenih gastroshiza prelazi 90%. U perinatologiji se postavljaju dva bitna pitanja: na koji način i kada je najbolje završiti ovakvu trudnoću? Ova pitanja predstavljaju predmet mnogih istraživanja.

U svojoj studiji, O'Connell i saradnici [26] preporučuju porođaj u 37. nedelji trudnoće, uz napomenu da carski rez ostaje rezervisan samo za trudnice koje imaju uobičajene indikacije za isti. Kada se beba rodi i smesti u jedinicu intenzivne nege, inicijalni terapijski ciljevi su održavanje homeostaze intravenskom nadoknadom tečnosti, respiratornom potporom, termoregulacijom i zaštitom creva [27]. Ovaj rad se bavi izučavanjem hirurškog tretmana gastroshize i povezanošću primenjenih hirurških tehnika sa pojmom komplikacija.

Optimalni hirurški pristup je i dalje tema mnogih istraživanja. Bez obzira na to koja se tehnika primenjuje, cilj hirurškog tretmana gastroshize je reponiranje creva i zatvaranje abdomena uz izbegavanje porasta intraabdominalnog pritiska iznad fizioloških vrednosti. Ovo se može postići neposredno nakon rođenja, primarnim manuelnim reponiranjem creva i suturiranjem fascijalnog defekta ili odloženim reponiranjem creva postavljanjem silastične vrećice i kasnjim zatvaranjem defekta. Primarno zatvaranje nije uvek moguće i bezbedno, posebno kada postoji abdominovisceralna disproportcija. Tada postoji mogućnost nastanka abdominalnog kompartment sindroma. Ova potencijalno letalna komplikacija zahteva neodložno otvaranje abdomena. Tako je *silo* tehnika u početku korišćena samo u slučajevima koji nisu bili podložni primarnom zatvaranju. Međutim, mnogi izvori zagovaraju primenu obe tehnike. Brojne studije ukazuju na to da je njihova uspešnost uglavnom izjednačena [28]. Novija istraživanja ističu značaj vremena proteklog od rođenja do operacije – optimalno je to manje od tri sata [29].

U našem studijskom uzorku je kod 62,3% (38/61) ispitanika primenjena tehnika primarnog reponiranja i fascijalnog zatvaranja defekta, a kod 37,7% (23/61) ispitanika je primenjena silastična vreća sa ojačanim prstenvom. Uočeno je da je učestalost nastanka NEC-a bila statistički značajno veća u SKG-u. Stopa učestalosti NEC-a je bila 21,7% (5/23) kod ispitanika u SKG-u, a kod ispitanika u HKG-u, NEC nije evidentiran. Apsolutni rizik od nastanka NEC-a u studijskom uzorku je bio 22% i bio je povezan sa tehnikom primene silastične vreće (u populaciji se AR se kretao od 7% – 44%). Rizik od nastanka NEC-a je bio 0,32 puta veći u SKG-u nego u HKG-u, sa 95%-im intervalom poverenja (RR 0,32, 95% CI: 0,22 – 0,47,  $p = 0,003$ ).

Naše istraživanje pokazuje da je primarno odloženo zatvaranje defekta povezano sa većim rizikom

troschisis cohort and the gastroschisis surgical cohort, as to the frequency of fatal outcome.

## DISCUSSION

Appropriate treatment of gastroschisis requires the integration of several disciplines (surgery, gynecology and obstetrics, neonatology, pediatrics). Gastroschisis can be diagnosed at regular follow-up ultrasound examinations during pregnancy, as well as by determining the mother's serum alpha-fetoprotein level (screening), which indicates the utmost importance of prenatal care. In developed countries, the number of gastroschisis cases detected in this way exceeds 90%. There are two important questions arising in perinatology: how and when is the best way to end this kind of pregnancy? These questions are the subject of many studies.

In their study, O'Connell et al. [26] recommend delivery at 37 weeks of pregnancy, noting that a cesarean section remains reserved only for pregnant women who have the usual indications for it. Once the baby is born and placed in the intensive care unit, the initial therapeutic goals are to maintain homeostasis through intravenous fluid replacement, respiratory support, thermoregulation, and bowel protection [27]. This study deals with the analysis of the surgical treatment of gastroschisis and the association between the applied surgical techniques and the occurrence of complications.

The optimal surgical approach is still the subject of many studies. Regardless of the technique used, the goal of the surgical treatment of gastroschisis is to reposition the intestine and close the abdomen while avoiding an increase in intra-abdominal pressure above physiological levels. This can be achieved immediately after birth by primary manual bowel repositioning and suturing of the fascial defect or with delayed bowel repositioning achieved by the placement of a silo bag and subsequent closure of the defect. Primary closure is not always possible and safe, especially when abdominovisceral disproportion exists, which is when there is a possibility of abdominal compartment syndrome. This potentially fatal complication requires immediate opening of the abdomen. Thus, the silo technique was initially used only in cases that were not amenable to primary closure. However, many sources advocate the use of both techniques. Numerous studies indicate that their success rate is generally equal [28]. Recent research emphasizes the importance of the time elapsed from birth to surgery – optimally it is less than three hours [29].

In our study sample, the technique of primary repositioning and fascial closure of the defect was applied in 62.3% (38/61) of the subjects, while a silo bag with a reinforced loop was used in 37.7% (23/61) of the patients. The incidence of NEC was statistically signifi-

od nastanka NEC-a, mada patofiziologija NEC-a povezanog sa tehnikom primene silastične vreće nije u potpunosti jasna. Iako bi pre mogle da se očekuju komplikacije ishemija i gangrena creva pri primeni ove hirurške metode u zbrinjavanju gastroshize, moguće je da je razlog i u tome što se ona možda češće primenjuje, usled male abdominalne šupljine, kod prevremeno rođene dece, koja su male telesne mase i sklona su razvoju NEC-a.

Takođe, rezultati našeg istraživanja sugerisu da pacijenti tretirani primarnim reponiranjem evisceriranih organa i fascijalnim zatvaranjem defekta mogu imati bolje ishode, jer su izbegli rizik od nastanka NEC-a. Nadalje, izbegavanje elektivnih preterminskih porođaja bi moglo da smanji učestaošću NEC-a kod pacijenata sa gastroshizom [30].

Za ostale komplikacije nije dokazana statistička značajnost razlike – ileus (RR 0,59, 95% CI: 0,47 – 0,73,  $p = 0,069$ ) i kompartmenat sindrom (RR 0,61, 95% CI: 0,50 – 0,75,  $p = 0,263$ ), iako je rizik od njihovog nastanka bio veći u hirurškoj kohorti gastroshiza pri primeni metode primarnog reponiranja evisceriranih organa i fascijalnog zatvaranja defekta, dok je rizik od nastanka smrtnog ishoda (RR 1,26, 95% CI: 0,46 – 3,43,  $p = 0,600$ ) bio veći u silastičnoj kohorti gastroshiza, pri primeni silo tehnike kod odloženg primarnog zbrinjavanja gastroshize.

I pored postojećih rezultata, teško je reći koja će metoda dati bolji ishod. Dok jedna metaanaliza ukazuje na to da primarno fascijalno zatvaranje daje bolje rezultate [16], druga pokazuje da bolesnici kod kojih je korišćena silastična vrećica ostaju kraće na mehaničkoj ventilaciji, ali imaju potrebu za dužom totalnom parenteralnom ishranom [31].

U radu iz 2020. godine Rejmonda i saradnika [32], može se pronaći podatak da je ukupno preživljavanje bolesnika sa gastroshizom do otpusta iz bolnice bilo neverovatnih 95%. Ipak, učestalost ponovljenih operacija nakon neonatalnog perioda je značajna, jer se približno 25% ovih bolesnika podvrgava hirurškom tretmanu tokom prve godine života, zbog intestinalne opstrukcije.

Visandži i saradnici [33] objašnjavaju da je Prognostički skor gastroshize (engl. *Gastroschisis Prognostic Score – GPS*) trenutno najbolje raspoloživo sredstvo za brzu i pouzdanu procenu težine bolesti. Ovaj sistem su, 2012. godine, kreirali Kouan i saradnici. Pomoću njega se može odrediti stepen izloženosti evisceriranih organa amnionskoj tečnosti, te proceniti metod izbora i ishod lečenja, samo na osnovu izgleda creva. Izračunavanje vrednosti GPS-a se mora obaviti odmah nakon rođenja, primenom standardizovanog vizuelnog sistema bodovanja, kojeg određuju četiri elementa: „sjaj-

cantly more frequent in SGC. The frequency rate of NEC was 21.7% (5/23) in subjects belonging to the SGC, while in subjects belonging to the GSC, no NEC was recorded. The absolute risk of NEC in the study sample was 22% and it was associated with the silo bag application technique (in the population, the AR ranged from 7% – 44%). The risk of developing NEC was 0.32 times greater in SGC than in GSC, with a 95% confidence interval (RR 0.32, 95% CI: 0.22 – 0.47,  $p = 0.003$ ).

Our study shows that delayed primary closure of the defect is associated with a higher risk of NEC, although the pathophysiology of NEC associated with the silastic bag technique is not entirely clear. Although complications of intestinal ischemia and gangrene could be expected when applying this surgical method in the treatment of gastroschisis, the reason may be that it is applied more often, due to the small abdominal cavity, in premature children, who are of small body weight and prone to developing NEC.

Also, the results of our study suggest that patients treated with primary repositioning of eviscerated organs and fascial closure of the defect may have better outcomes because they have avoided the risk of NEC. Furthermore, avoiding elective preterm deliveries could reduce the incidence of NEC in patients with gastroschisis [30].

For other complications, the statistical significance of the difference was not proven – ileus (RR 0.59, 95% CI: 0.47 - 0.73,  $p = 0.069$ ) and compartment syndrome (RR 0.61, 95% CI: 0.50 - 0.75,  $p = 0.263$ ), although the risk of their occurrence was higher in the gastroschisis surgical cohort when applying the method of primary repositioning of eviscerated organs and fascial closure of the defect, while the risk of fatal outcome (RR 1.26, 95% CI: 0.46 - 3.43,  $p = 0.600$ ) was higher in the silastic gastroschisis cohort, when applying the silo technique in delayed primary care of gastroschisis.

Despite the existing results, it is difficult to say which method will give a better outcome. While one meta-analysis indicates that primary fascial closure gives better results [16], another shows that patients in whom a silo bag is used remain on mechanical ventilation for a shorter period, but require longer total parenteral nutrition [31].

In a 2020 paper by Raymond et al. [32], it was reported that the overall survival of patients with gastroschisis until discharge from hospital was an incredible 95%. However, the frequency of reoperations after the neonatal period is significant, because approximately 25% of these patients undergo surgical treatment during the first year of life, due to intestinal obstruction.

Wissanji et al. [33] explain that the Gastroschisis Prognostic Score (GPS) is currently the best available tool for a quick and reliable assessment of the severity of

na" creva, prisustvo nekroze, atrezije i/ili perforacije. Bolesnici se mogu svrstati u dve kategorije: pacijenti sa malim rizikom ( $GPS \leq 1$ ) i pacijenti sa velikim rizikom ( $GPS \geq 2$ ) [34,35].

Ukoliko se procenom izgleda uoči prisutan *peel* na crevima, a pored toga postoji i abdominovisceralna disproporcija, ovo predstavlja indikaciju za postavljanje silastične vrećice [36]. Iako su indikacije jasne, postavljanju silastične vrećice treba pristupiti oprezno, jer ukoliko je otvor na prednjem trbušnom zidu premali (< 1,5 cm), vaskularizacija može biti dodatno kompromitovana i dovesti do ishemije creva i daljih komplikacija. Zato je na hirurgu da proceni da li je potrebno načiniti dodatnu fasciotomiju pre postavljanja silastične vrećice, kako bi otvor na prednjem trbušnom zidu bio adekvatne veličine. Upravo kod ove procene može doći do greške, što samim tim i predstavlja razlog češće udruženosti NEC-a i smrtnog ishoda sa primenom silastične vrećice u lečenju gastroshize.

U našem istraživanju su odgovori na mnoga pitanja ostali nedorečeni. Razlog tome je i mali uzorak ispitanih, pa samo istraživanje nema veliku statističku snagu. Veličina uzorka je u skladu sa podatkom da je gastroshiza retka kongenitalna anomalija.

## ZAKLJUČAK

Ne postoji jedinstveni dokaz o superiornosti jedne metode u lečenju gastroshize nad drugom. Istraživanjem je utvrđeno da su rizici od nastanka ileusa i kompartment sindroma veći pri primeni tehnike primarnog fascijalnog zatvaranja defekta, a da je rizik od nastanka NEC-a i smrtnog ishoda veći prilikom primene silo tehnike. Izbor metode koja će biti primenjena u lečenju zavisi od prisustva ili odsustva abdominovisceralne disproporcije i izgleda evisceriranih creva, procenjenih primenom GPS-a. Iz dosadašnjih studija se može reći da je uspeh u lečenju gastroshize veliki, uz posebno obraćanje pažnje na perioperativni tretman i hiruršku tehniku. Ipak, potrebne su buduće multicentrične studije radi sistemskog pregleda i statistički značajnih rezultata.

**Sukob interesa:** Nije prijavljen.

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the disease. This system was created in 2012 by Cowan et al. It can be used to determine the degree of exposure of eviscerated organs to amniotic fluid, as well as to evaluate the method of choice and the outcome of treatment, based only on the appearance of the intestine. GPS scoring and calculation must be done immediately after birth, using a standardized visual scoring system, which is determined by four elements: 'glossy' intestines, the presence of necrosis, atresia, and/or perforation. Patients can be classified into two categories: low-risk patients ( $GPS \leq 1$ ) and high-risk patients ( $GPS \geq 2$ ) [34,35].

If the assessment of the appearance of the intestines shows the presence of a 'peel', i.e., 'rind' on the intestines, and there is also an abdominovisceral disproportion, this is an indication for the placement of a silastic bag [36]. Although the indications are clear, the placement of the silastic bag should be approached with caution, because if the opening on the anterior abdominal wall is too small (< 1.5 cm), the vascularization may be further compromised and lead to intestinal ischemia and further complications. Therefore, it is up to the surgeon to assess whether it is necessary to perform an additional fasciotomy before placing the silo bag so that the opening on the anterior abdominal wall is of the appropriate size. It is precisely in this assessment that an error may occur, which is why there is a more frequent association of NEC and fatal outcome with the use of a silo bag in the treatment of gastroschisis.

In our study, the answers to many questions remain unanswered. The reason for this is the small sample of respondents, which is why the research itself does not have great statistical power. The size of the sample is consistent with the fact that gastroschisis is a rare congenital anomaly.

## CONCLUSION

There is no distinctive proof of the superiority of one method in the treatment of gastroschisis over another. The study found that the risks of ileus and compartment syndrome are higher when the technique of primary fascial closure of the defect is applied, as well as that the risk of NEC and fatal outcome is higher when using the silo method. The choice of the treatment method depends on the presence or absence of abdominovisceral disproportion and the physical appearance of the eviscerated intestines, assessed with GPS. Based on the studies published so far, it can be said that success in the treatment of gastroschisis is high and that special attention needs to be paid to perioperative treatment and surgical technique. However, future multicentric studies are necessary for a systematic review and statistically significant results.

**Conflict of interest:** None declared.

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