The aim of the study was to assess the value of the today’s appropriate approach, preterm delivery in the 34th week of gestation by Caesarean section and subsequent surgical intervention at the perinatal center, in daily practice of pediatric surgery with regard to early postoperative and mid-term outcome.

Material and methods. Over the time period of 9 years, all consecutive cases diagnosed with gastroschisis at the perinatal center, University Hospital of Magdeburg, were born by Caesarean section within the 34th week of gestation followed by surgical intervention. The registered data were compared with those published by other groups.

Results. Overall, there were 19 cases through the investigation period from 01/01/2006 to 12/31/2014. The mean duration of gestation was 237.9 days. The mean birth weight was 2,276 g. In all individuals, a primary closure with no artificial material was achieved. The duration of postoperative artificial respiration was 2.3 days. Oral uptake could be initiated on the 10th postoperative day on average. The mean hospital stay was 37 days. There was no lethality. As complications, postoperative (iv catheter-associated) sepsis occurred in one case and relaparotomy became necessary in a further case because of no possible completion of enteral nutrition by 20 days after primary closure (complication and relaparotomy rate, 10.5% and 5.26%, respectively).

Conclusions. The data indicate that in case of gastroschisis, primary closure can be more frequently achieved by section within the 34th week of gestation. Under the prediction of an optimal neonatological care, the risks of a preterm delivery by a planned section appear to be manageable.

Key words: prevalence data, gastroschisis, preterm delivery, caesarean section
Surgical treatment results in gastroschisis

by Caesarean section and subsequent surgical intervention at a perinatal comprehensive center in case of an already prenatally known gastroschisis. Since the pathogenetic background indicates changes of the intestine by intrauterine contact with amniotic fluid over the last 6 weeks of pregnancy, the approach can only be successful in case of gastroschisis.

MATERIAL AND METHODS

History

The first scientific report on gastroschisis can be assigned to Calder in 1733 (1). In 1878, Fear achieved a successful treatment of gastroschisis for the first time (2). The therapeutic choice, in particular, with regard to the specific procedure depends on the extent of the local finding and the disproportion between prolapsed visceral organs and the volume of the peritoneal cavity. In repositioning, an increased intraabdominal pressure is generated, which affects the amplitude of diaphragmatic excursions. This leads to an aggravation of the artificial respiration (3) and/or, in case of an increase of the airway pressure, to a relevant pathological aberration of important intraabdominal parameters, which can induce a relative or absolute mesenteric ischemia (4). The first preventative approach comprised creation of an “artificial hernia“ (only skin closure according to the technique by Gross (5). Therefore, Schuster inaugurated a technique using synthetic material (6). Alternatively, enlargement and closure of the peritoneal cavity was achieved by the means of a patch plasty through the following years. The search for biocompatible concepts is still ongoing and can be characterized as being under clinical testing – therefore, a final conclusion cannot be made so far (7).

As an alternative approach, a modification as “spring-loaded silo“ is currently widely discussed or the procedure by Bianchi is considered a true alternative to the “impossible“ primary closure (8, 9). In 2003, Jona described the “Gentle Touch Technique“ (10). To which extent an additional pull at the silo can lead to a faster reposition, cannot be satisfyingly assessed presently (11). As a further option, Werbeck et Koltai favor temporary coverage with umbilical cord (12). In 2013, Gomes et al. reported their first experiences of the use of the Alexis wound retractor (13). In summary of all experiences, it can be stated that primary closure needs to be preferred despite all the improvements of materials and surgical techniques (fig. 2).

Etiology and pathogenesis

The etiology of gastroschisis in detail is not exactly clarified yet. It has been suggested a polygenic multifactorial genesis. According to novel hypotheses, a pressure-like symptomatology is assumed at the differentiating abdominal wall leading, as a consequence of this,
to an initial tearing at the weakest site of it (near the right umbilical vein) followed by excrescence of the intestine (14). This mechanism would also allow to explain left-sided occurrence of gastroschisis (15) in cases of which the left vein can be considered a weak point of stability (16). How far the additional occurrence of a coagulation disorder can provide significant impact (17), has not been completely elucidated yet. During the development of an experimental approach using embryos of chicks, Tibboel et al. were able to demonstrate in 1985 that occurring intestinal changes need to be considered consequences of the unphysiological fluid composition of the intestine’s surroundings (18). An initial causative disorganization of the nerval structure of the abdominal wall was definitely denied.

Prevalence and malformation registry

Prevalence is defined as frequency of a disease related to a certain population group and needs to be differentiated from incidence, which is defined as number of a new disease related to a formerly healthy population. Current international data on the prevalence of congenital defects of the abdominal wall were obtained from various cooperating malformation registries, which pursue the standardized documentation of malformations and, thus, allow to compare their data among each other (19). Internationally, ICBDSR (International Clearinghouse for Birth Defects Surveillance and Research with 46 centres worldwide) and EUROCAT (European Surveillance of Congenital Anomalies with 49 members from 20 European countries) are the most important registries. The population-based-birth defects registry of the German district Saxony-Anhalt (Malformation Monitoring Centre Saxony-Anhalt) located at the Medical School Otto-von-Guericke University of Magdeburg (Germany) became the 33rd member of EUROCAT in 2002 (20). For the last years, an increased frequency of gastroschisis by trend has been reported worldwide (21, 22, 23). However, this cannot be confirmed for Saxony-Anhalt (fig. 3). Looking at all centers registered in EUROCAT, there is a slight increase of the prevalence of gastroschisis detectable.

Prenatal diagnostics

The first description of gastroschisis was most likely provided by Donald in 1958 (24). In Germany, there have been three transabdominal ultrasound-based investigations mandatory by law during pregnancy since 1995. From the 12th week of gestation on, hints of ventral defects of the abdominal wall can become detectable since until this time point, physiological umbilical hernia exists (fig. 4). Today, the supervision of this gravidity at a center of perinatal medicine appears to be obligatory. It cannot be estimated whether it can be achieved using ultrasound-based criteria to assess postnatal prognosis (25).

Neonatology and anesthesiological aspects

Beside the intervention-specific difficulties, physiological and pathophysiological aspects may not be disregarded in surgical interven-

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Fig. 3. Prevalence of gastroschisis in Europe and the German district Saxony-Anhalt from 1987 to 2012

Fig. 4. Representative transabdominal ultrasound image – gastroschisis within 23rd week of gestation
Surgical treatment results in gastroschisis

tions of premature infants and newborns (26). Development of a bronchopulmonary dysplasia (BPD) and its long-term consequences (27) cannot always be precluded even with modern methods of artificial respiration (such as high frequency) (28). In addition to the age-specific maturation of the lung, the very relevant diaphragmatic impulsion of the respiration can be severely affected in abdominal surgery. This is, in particular, valid in surgical interventions which induce an increase of the former pressure (e.g., hernia of the diaphragm, omphalocele, gastroschisis). Furthermore, maintenance of the extraterine circulation needs to be particularly observed. By persisting ductus arteriosus (29) or foramen ovale, shunt-like collaterals can develop reflecting the intrauterine situation, which need to be preoperatively diagnosed. One of the greatest dangers is the formation of a periventricular bleeding (IVH), which is directly influenced by the duration of gestation (30) and various other factors (31). Similar aspects can be reported on the periventricular leucomalacia (PVL). All surgical interventions in this area require an extreme sensibility (32, 33), which determines considerably the long-term outcome in the treatment of premature infants. For instance, retinopathy of the premature infant (34) can be attenuated or even prevented in many cases. Furthermore, thermoregulation, infusion therapy and nutrition (35-38) need to be paid attention. In a procedure, which plans terminated Cesarean section within the 34th week of gestation, risks need to be appropriately assessed, e.g., with regard to mortality of such a premature infant, for which newborn registries such as that of the German district Saxony-Anhalt can serve. So, there has been no different mortality of premature infants (32nd to 36th week of gestation) vs. newborns delivered after the 36th week of gestation. This registry includes all infants who were treated in a department of pediatrics after birth (tab. 1).

Modus and time point of delivery

There has been a still ongoing discussion on the best modus of delivery for a long time (39, 40); however, it has not been objectively, with no further doubt and satisfyingly, clarified yet (41, 42). For the appropriate time point of delivery, the induced maturation of the lungs plays a substantial role in the immediate prenatal time frame since only this maturation allows an iatrogenically induced preterm delivery by Cesarean section even prior to the 36th week of gestation. Basically, normal physiological sequences are guaranteed by the passage through the natural birth canal, which can only severely mimicked by the iatrogenically induced delivery. On the other hand, almost not calculable or assessable mechanic and infectious aspects can occur by the compression during this passage, which can lead to undesirable consequences in case of external abnormalities or structures under pressure.

Table 1. Mortality depending on the week of gestation (Newborn Registry of the German district Saxony-Anhalt from 2000 to 2009)

<table>
<thead>
<tr>
<th>Jahr</th>
<th>Week of gestation</th>
<th>In total (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>prior to 26 (%)</td>
<td>26-27 26-28 * (%)</td>
</tr>
<tr>
<td>2000</td>
<td>35,7</td>
<td>18,9 (%)</td>
</tr>
<tr>
<td>2001</td>
<td>43,6</td>
<td>4,8 (%)</td>
</tr>
<tr>
<td>2002</td>
<td>40,7</td>
<td>12,8 (%)</td>
</tr>
<tr>
<td>2003</td>
<td>45,5</td>
<td>13,7 (%)</td>
</tr>
<tr>
<td>2004</td>
<td>64,9</td>
<td>6,1 (%)</td>
</tr>
<tr>
<td>2005</td>
<td>45,9</td>
<td>16,7 (%)</td>
</tr>
<tr>
<td>2006*</td>
<td>50,0</td>
<td>12,7 (%)</td>
</tr>
<tr>
<td>2007*</td>
<td>41,9</td>
<td>9,7 (%)</td>
</tr>
<tr>
<td>2008*</td>
<td>39,1</td>
<td>8,2 (%)</td>
</tr>
<tr>
<td>2009*</td>
<td>36,8</td>
<td>16,7 (%)</td>
</tr>
</tbody>
</table>

* Modification of the time frame (periods) since 2006; since 2010, there has been no further general differentiation of mortality later than the 32nd week of gestation.
For gastroschisis, Swartz et al. (1986) recommended a differentiated approach including the various options and were, thus, able to achieve a distinct reduction of mortality (43). Hagberg et al. referred to the slight intestinal adhesions within the 38th week of gestation and favored the elective Caesarean section at this time point (44). Subsequently, Swift and colleagues (45) recommended in 1992 the planned Caesarean section within the 37th week of gestation, which was also pursued by other authors (46) and which was related to observations and recommendations by Moore (47). In contrast, Logghe et al. did not detect any positive impact onto the outcome within the 36th week of gestation (48). In 2010, Henrich et al. (49) advised no primary section in case of a fetus with malformation of brain, heart, urogenital tract and skeleton whereas in a fetus with gastroschisis, omphalocele or spina bifida, they found an advantage related to logistics and avoidance of a laceration and denied significant long-term advantageous effects.

Overall, there seems to be a trend favoring the planned section in the literature including a further positive development in the same direction even in centers with a more aggressive opinion with regard to natural delivery (50, 51). In addition to the modus of delivery, the time point is considered a further crucial aspect, which is far better controllable in Caesarean section. Based on this, it could be assumed that a distinct improvement of the outcome would have become evident but recent publications by Carroll et al., Soares et al. or Reigstad et al. did not provide such evidence (52, 53, 54).

RESULTS

Over the time period from 01/01/2006 to 12/31/2014, there were 21 cases of gastroschisis, two of them were excluded, in particular. One case was born within the 27/3 week of gestation because of a presumed perforation of the extended gut. The birth weight was 1,080 g, an atresia of the colon was found and primary closure of gastroschisis was possible with no need for patch material. The child survived with primary healing and colostoma in situ; finally, the patient was discharged on the 71st postoperative day.

The second case was born outside within the 36th week of gestation. The birth weight was 2,240 g. No intestinal atresia was found and primary closure became possible too. The child survived with primary healing and could be discharged on the 26th day after birth.

Thus, there were 19 cases, with a prenatally detected gastroschisis born by sectio caesaarea in our hospital.

The mean gestational age was 34 weeks (= 237.9 d; SD, 5.0 d).

The mean birth weight was 2,276.8 g (range, 1,750-2,900 g).

There were 7 girls and 12 boys. The mean age of the mother was 21.9 (range, 18-29) years.

The mean time from Caesarean section to operation was 3 (range, 01:05-04:30) hours.

A primary closure was achieved in all cases, with no need for a patch or silo.

The duration of postoperative artificial respiration was 2.3 d in average (range, 1-7 d).

First oral nutrition was given on day 10 postinterventionally (range, 6-25 d).

The discharge from the hospital became possible on day 37 in average (range, 23rd-101st d).

In one case, relaparotomy was required because of unsuccessful completion of total oral nutrition on day 20 (relaparotomy rate, 5.26%; n=1/19). In a further case, venous catheter-associated sepsis occurred (complication rate, 10.5%; n=2/19). Other complications such as intracerebral bleeding or pneumothorax by the preterm section or further specific (associated to the surgical intervention) or general complications were not observed. There was no death.

DISCUSSION

Comparing data from other authors, it can be stated that in the majority of cases the birth weights of infants with gastroschisis are higher than observed in the own case series. For instance, Snyder reported a mean birth weight of 2,501 g for infants with gastroschisis within the 36th week of gestation whereas Tunell et al. published a mean birth weight of 2,500 g for the decade from 1975-1984 (55, 56). In addition, Davis et al. described a mean birth weight of 2,400 g with SD of 600 g in 46 cases with gastroschisis from 1998-2007 (57). In
contrast, Haddock et al. found a mean birth weight of 2,170 g (35.8th week of gestation) in 1996 analyzing data from 1983-1993 (58).

In 2012, Safavi et al. listed 507 cases with gastroschisis in Canada from 2005-2012 with a mean duration of gestation of 36.1 weeks and a mean birth weight of 2,541 g (59). It turned out that planned induction of labour within the 34th week of gestation by Caesarean section does not result in diminution of the birth weight. This may allow to conclude that from this time point, no further increase of the body weight can be expected since this process is affected by the initial peritonitis(-like) changes. To demonstrate this, we show the mean birth weights over the recorded cases in our hospital in tab. 2.

The reported results by Moir et al., Vegunta et al., Gelas et al., Hadidi et al. and Serra et al. as listed in tab. 3 confirm this too (60-64).

The mean age of the mothers with gastroschisis infants was 23.91 (±5.86) years from 1997-2005 and 21.89 (±3.10) years from 2006-2014, respectively. In several publications, younger age of the mother is associated with an increased prevalence of gastroschisis and an older age with an increased frequency of omphalocele, respectively (65, 66, 67). Even the age of the father may have an impact onto the occurrence of gastroschisis (68). In 2007, Loane described that – based on the analysis of 936 cases with gastroschisis in 15 European countries from 1980-2002 – increasing prevalence cannot only be explained by the younger age of the mothers, e.g., Materna-Kiryuk et al. could not find such association in Poland (69, 70). Contrarily, Gill et al. calculated a significant higher risk for
– the infant to be born, and
– mothers younger than 20 years of age to deliver a baby
with gastroschisis in the U.S. from 1997-2007 (OR, 6.1; 95% CI, 4.8-8-0) (71). A recent investigation by Getz et al. suggests that a short time period between two gestations may also promote the occurrence of gastroschisis (72). In addition, food pattern and smoking are important aspects, which need to be included into considerations on the age of the mothers for assessing the risk of gastroschisis (73, 74, 75). Furthermore, the younger age seems to correlate with a low education level, which may have direct impact onto food pattern, smoking and drug abuse. Therefore, the only consideration of the mother’s age appears not to be reasonable. Because of the fact that in case of gastroschisis, an elective Caesarean section within the 34th week of gestation has been performed since 2006, this should have led to an expectable shortening of gestation. However, the data indicate only a shortening by 10 days in average and, therefore, can be considered a significant aspect but with no disadvantageous impact. All born infants with gastro-

<table>
<thead>
<tr>
<th>Time period</th>
<th>Birth weight of gastroschisis patients (g)</th>
<th>Gastrochisis (n)</th>
</tr>
</thead>
<tbody>
<tr>
<td>1960–1980</td>
<td>2109</td>
<td>21</td>
</tr>
<tr>
<td>1981–1985</td>
<td>2317</td>
<td>7</td>
</tr>
<tr>
<td>1986–1990</td>
<td>2400</td>
<td>7</td>
</tr>
<tr>
<td>1991–1995</td>
<td>2281</td>
<td>6</td>
</tr>
<tr>
<td>1996–2000</td>
<td>2272</td>
<td>5</td>
</tr>
<tr>
<td>2001–2005</td>
<td>2235</td>
<td>9</td>
</tr>
<tr>
<td>2006–2014</td>
<td>2276</td>
<td>19</td>
</tr>
</tbody>
</table>

Table 3. List of main criteria with direct comparison of the results of various authors and study groups

<table>
<thead>
<tr>
<th></th>
<th></th>
<th></th>
<th></th>
<th></th>
<th></th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>No. of patients</td>
<td>13</td>
<td>30</td>
<td>13</td>
<td>23</td>
<td>36</td>
<td>19</td>
</tr>
<tr>
<td>Week of gestation</td>
<td>34,2</td>
<td>35,7</td>
<td>34,7</td>
<td>&lt; 36</td>
<td>33,5</td>
<td>34</td>
</tr>
<tr>
<td>Birth weight (g)</td>
<td>2307</td>
<td>2275</td>
<td>2250</td>
<td>2079</td>
<td>1940</td>
<td>2276</td>
</tr>
<tr>
<td>Primary closure rate(%)</td>
<td>100</td>
<td>83</td>
<td>100</td>
<td>87</td>
<td>87</td>
<td>100</td>
</tr>
<tr>
<td>Duration of artificial respiration (days)</td>
<td>1,3 day</td>
<td>3 days</td>
<td>2 days</td>
<td>3,8 days</td>
<td>2,3 days</td>
<td></td>
</tr>
<tr>
<td>Mortality (%)</td>
<td>0</td>
<td>10</td>
<td>0</td>
<td>13,5</td>
<td>0</td>
<td>13,5</td>
</tr>
<tr>
<td>p.o. nutrition (days)</td>
<td>19,1</td>
<td>19</td>
<td>4</td>
<td>13,5</td>
<td>13,5</td>
<td>13,5</td>
</tr>
<tr>
<td>Hospital stay (days)</td>
<td>22,7</td>
<td>24</td>
<td>22,5</td>
<td>51,6</td>
<td>58,5</td>
<td>37</td>
</tr>
</tbody>
</table>
schisis after 2006 underwent primary closure with no artificial material, which can be considered a substantial improvement compared with previous years or time periods. The mean duration of the artificial respiration until 2006 was 7 days whereas from 2006 to 2014, the duration was 2.3 days only.

Because of gaps in continuous data acquisition until 2006, in particular, because of great advances and several novel aspects in the care of newborns (which had not been documented before), it seemed to be not useful or allowed to try to create a statistical evidence.

The approach of an elective Caesarean section within the 34th week of gestation in case of prenatally known gastroschisis can be considered advantageous based on the fact that the duration of the postoperative artificial respiration could be decreased. The uptake of fluid and nutrition was distinctly earlier after 2006 (by 10 days) than until 2006 (by 21 days), the hospital stay was shorter from 2006 on (37 days) vs. until 2006 (68 days).

Because of the little groups and the changes in technical and medical advantages it is not really clear, how big the single influence of the preterm delivery is.

Between 1960 and 1970, all infants with gastroschisis died – in contrast, from 2001 to 2014, all infants with gastroschisis survived (fig. 5).

This favorable development was caused by the improved technical conditions, e.g., in 1981, Hagberg et al. referred to the influence of the advances of neonatology, changes in the logistics and the improved preoperative conditions of the infants with gastroschisis and their local findings, which need to be treated and undergo surgical intervention (76). This can be confirmed by data from Moir et al., Vegunta et al., Gelas et al., Hadidi et al. and Serra et al. (60-64) (tab. 3). The data show a range from 0 to 10 %.

In 1988, Moore suggested the approach of an “elective preterm section” for the treatment of infants with gastroschisis for the first time based on 2 impressive case reports (30th and 36th week of gestation) (47). He had researched on the topic since the early fifties (77) and published a number of scientific papers on it (78-84). Hagberg et al. confirmed this idea and approach in the “Zeitschrift für Kinderchirurgie” in the same year based on 7 own cases (38th week of gestation) (44). So, Moore can be considered the “Nestor” of the basic approach as described above. But only the development of neonatology, in particular, the advances in respiration have provided the breakthrough.

While during the first years, only a few scientific articles on the topic were published (85-88), there has been a distinct trend in the gastroschisis treatment since 2000 though there were also single publications with no positive impact (48). But, in particular, the results by Moir et al., Vegunta et al., Gelas et al., Hadidi et al. and Serra et al. provided a broad scientific base (60-64). The reported results can also impressively confirm their data.

### SUMMARY

Cases with gastroschisis seem to rise since by an early diagnosis, the basically good prognosis and a close-mesh care at perinatal centers do seldomly lead to abortion. The elective preterm Caesarean section (within the 34th week of gestation) as a favorable factor from a prognostic perspective can be considered as evident (fig. 6A and 6B).

However, the amnion-exchange procedure has not – so far – provided evidence for a definitive positive effect in cases with gastroschisis. Fetal surgery appears – despite an impressive development and technical specification – not justified in a survival probability of more than 90%. According to a questioning of EUP-
Surgical treatment results in gastroschisis

SA delegates (European Paediatric Surgeons Association) what approach is favored in pre-natally known gastroschisis, Zani found out in 2012 that the non-elective delivery belongs to the normal logistic in Western Europe with 46% and Eastern Europe with 58% (89). This type of approach should be critically discussed.

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