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Incidence of perinatal complications in children with premature craniosynostosis

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Abstract

Aims: To study the incidence of perinatal complications in children diagnosed with premature craniosynostosis and their mothers.
Methods: Questionnaires were sent to all women (n = 220) who delivered a child diagnosed either pre- or postnatally with craniosynostosis and treated at the University Hospital of Innsbruck, Austria between January 1, 1990 and October 10, 2007. The incidence of various complications in the group of children with craniosynostosis was compared with data from the Birth Registry of Tyrol, which served as a comparison population in this study (n = 57,317).
Results: A total of 46.4% of the questionnaires were returned (n = 102). Children diagnosed with craniosynostosis showed a significantly higher rate of fetal malpresentations at birth [P < 0.001; OR 2.38 (CI 1.53–3.70)] compared to the general population. In particular, the rate of abnormal cephalic [P < 0.001; OR 3.42 (CI 2.03–5.76)] and breech presentations [P = 0.01; OR 2.39 (CI 1.27–4.49)] was significantly increased. In 10% of all neonates the Apgar score (P = 0.001) as well as the pH-value (P < 0.001) was found to be at least one category lower than in the comparison population.
Conclusions: Children diagnosed with craniosynostosis have a significantly higher rate of numerous birth complications compared to the overall births documented at the Birth Registry of Tyrol.

Keywords: Craniosynostosis; fetal malpresentation; perinatal complication; postnatal complication.

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Introduction

Craniosynostosis, the early fusion of one or more skull sutures, is a major malformation with a prevalence of ~1 in 2100–3000 children [8]. It represents a serious abnormality in infancy and childhood requiring proper diagnosis and treatment. The premature fusion of cranial sutures can occur alone (isolated) or together with other anomalies, resulting in one out of 150 different possible syndromes [3, 4, 25].

Cranial sutures have many functions, and are intricately involved in the development and growth of the expanding cranial bones [19]. Furthermore, cranial sutures also play an essential role during vaginal delivery. At birth, areas where sutures will develop permit adjusted overlap of the calvarial bones as the fetal head becomes compressed during passage through the birth canal [11, 25]. The resulting molding of the neonatal skull normalizes during the first week of life by cranial re-expansion and widening of sutural areas [4].

Considering the fundamental role of cranial sutures in allowing for the safe passage of the fetal head through the maternal birth canal and single reports about various complications in craniosynostoses at birth [1], the premature suture fusion may lead to an increased rate of perinatal complications in affected children and their mothers. Previous studies have only focused on perinatal information in deformational plagiocephaly, which is an abnormal head shape caused by external pressure [16]; however, to our knowledge, no study to date examined this issue in craniosynostoses (MEDLINE; 1966 to May 2009; English language; search terms ''perinatal complications'', ''postnatal complications'' and ''craniosynostosis''), which is caused by premature suture fusion. This is also of increasing importance given the major advances in prenatal diagnosis that allow for the definite antenatal diagnosis of craniosynostosis using ultrasound and MRI [5, 6, 17]. Therefore, the purpose of the present study is to clarify the incidence of potentially occurring perinatal complications in children with craniosynostosis, as well as to raise the awareness of possible prophylactic antenatal measures.

Materials and methods

This retrospective study was carried out after approval from the Institutional Review Board of the Birth Registry Austria in 2008. All infants with premature craniosynostosis born between January 1, 1990 and October 10, 2007 who were diagnosed and treated at the Clinical Department of Plastic and Reconstructive Surgery (University Hospital) in Innsbruck, Austria were eligible for the study (n = 220). This department is a multidisciplinary reference center for craniosynostosis patients in the western part of Austria, implying

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that all craniosynostosis cases in this area were referred to this institution by primary health care providers during the study period. The inclusion criteria were set in order to provide an adequate study population that is representative of craniosynostosis cases in general. For the statistical comparison with the general population, affected children born between 2000 and 2007 were extracted from the entire craniosynostosis group (n = 51). Perinatal complications in the study group were compared to the data provided by the Birth Registry of Tyrol [18], an evaluation of all births (n = 57,317) in Tyrol between 2000 and 2007, and the results were analyzed retrospectively. Under the hypothesis of a constant time trend within the birth registry data, indicated by a significant linear trend test in specific parameters of the registry data over years, a comparison of the registry data to the entire craniosynostosis population (1990–2007; n = 102) was performed to maximize the generalization of the results for children with premature craniosynostosis.

We used the Power Chart® – registry, a database that has tracked patients' diagnoses at this department since 1997, to identify craniosynostosis cases (Power Chart® Electronic Medical Record System, Cerner Inc., Kansas City, MO, USA). In patients that were born before 1997 the data were obtained from the internal patient-database (Filemaker Software®, Filemaker Inc., Santa Clara, CA, USA). We also queried the Mirror® digital imaging system, which contains pictures of all patients pre- and postoperatively, to identify cases that may have been missed inadvertently by the registry (Mirror® Cosmetic Surgery Digital Imaging, Canfield Scientific Inc., Fairfield, NJ, USA). Our case definition specified that the infant must have been diagnosed with premature craniosynostosis by an Innsbruck University Hospital health care provider. Children diagnosed with secondary deformational plagiocephaly or other non-synostotic malformations of the neuro- and viscerocranium were excluded from this study.

The study was based on information obtained from a questionnaire that was handed or sent via mail to all mothers (n = 220) of children that met the inclusion criteria in November 2007. The questionnaire consisted of 14 pages and the questions referred to data of the Austrian Mother-Child Pass in order to provide mothers with a standardized source of information. The Austrian Mother-Child Pass is a pregnancy and early-childhood medical record book where the health care provider documents all the relevant medical information about the mother and her child, perinatal complications as well as results of pre- and postnatal screening examinations. In this study, the Mother-Child Pass of craniosynostosis patients was used to obtain valid information concerning sophisticated pre-, peri- and postnatal parameters, such as fetal presentation, pH-value, Apgar-score or mode of delivery. A letter containing information and instructions about the questionnaire was also enclosed.

A total of 46.4% of the questionnaires were returned by March 2008 (n = 102) and no additional mailing was performed for non-responders. The demographic comparison concerning the date of birth, origin and craniofacial malformation of the group of responders and non-responders of the questionnaire showed no significant disparities. In consideration of the population-based approach of the current study and the high number of cases, the response rate was adequate to ensure that the results were representative of the target population of premature craniosynostoses.

Cases were categorized into two groups based on cesarean or vaginal mode of delivery. For the evaluation of particular questions the two groups were further subdivided into different entities of craniosynostoses along the classification of premature craniosynostosis by Marchac and Rénier [14].

Percentages were used to describe the incidence of various complications. Odds ratio (OR) was calculated for some criteria to provide an indicator of the potential probability of certain complications in the craniosynostosis group and respective 95% confidence intervals (95% CIs) were calculated. Statistical comparisons were performed with contingency table analyses (χ^2- or Fisher exact test as appropriate). Calculations were performed with the use of SPSS 15.0 (SPSS, Chicago, IL, USA) and GraphPad Quickcalcs (Graphpad Software, Inc., San Diego, CA, USA). Due to the high number of patients in the comparison group the *Yates' correction for continuity* was applied when appropriate to the χ^2-testings. All P-values were two-tailed and a $P < 0.05$ was considered statistically significant.

Results

One hundred singleton and two multiple pregnancies of children with premature craniosynostosis met the inclusion criteria and responded to the questionnaire (n = 102), of which 94.1% required surgical treatment during infancy. All had a positive clinical exam and radiographic findings consistent with premature craniosynostosis. The comparison group comprised of 56,475 singleton and 842 multiple pregnancies (n = 57,317).

In the group of mothers of children with craniosynostosis the median *duration of pregnancy* was 39.09 weeks (SD = 2.49). The rate of premature infants in this group was 11.8%, not significantly different than the overall births in Tyrol ($P = 0.65$). In 80.4% of the cases, mothers of children with craniosynostosis delivered between the 37th and 41st (37^{+0}–41^{+6}) week of gestation, 9.6% less than the rate in the comparison group (80.4%; $P = 0.002$). The percentage of patients having prolonged pregnancies (7.8%) was significantly higher in the group of craniosynostoses (1.2%; $P = 0.001$, Table 1).

In children with premature craniosynostosis the overall *birth weight* corresponded to the data from the Tyrolean comparison population. The rate of *induction of labor* preceding delivery was 38.5% of all craniosynostosis patients, which is significantly higher than in the comparison population [13.1%; $P < 0.001$; OR 4.15 (95% CI 2.63–6.55); Table 1]. Of these deliveries, labor was medically induced in 20.5%, in 14.1% an amniotomy was performed and both interventions were applied in 3.9%. Of the 61.5% spontaneous onset vaginal deliveries, 15.4% were born after premature rupture of the membranes (PROM) at term, 3.9% after preterm PROM and 10.3% by vacuum extraction. Only 29.5% of all vaginal births in the group of craniosynostoses showed a spontaneous vaginal delivery.

As many as 77.5% of all children with premature craniosynostosis were born by a vaginal *delivery*: 19.0% required additional measures like breech extraction or operative vaginal delivery. Vacuum extraction was performed in 11.4% and forceps delivery in 1.3% of cases. In 22.5% of craniosynostosis patients a cesarean delivery was performed. Compared to the Tyrolean population no significant differences were found between the mode of delivery (all $P > 0.05$; Table 1) except the rate of vaginal breech deliveries in mothers of children with craniosynostosis (3.9% vs. 0.2%; $P < 0.001$).

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Table 1 Rates of perinatal complications and parameters in craniosynostosis patients and the average population in (%).

Characteristics	Craniosynostoses 1990–2007 (n = 102)	Birth registry Tyrol 2000–2007 (n = 57,317)	P-value
Duration of pregnancy			
On-term deliveries	80.4	90.1	0.002**
Premature infants	11.8	9.9	0.648
Prolonged pregnancies	7.8	1.2	0.001**
Induction of labor	38.5	13.1	<0.001**
Fetal malpresentations	31.5	14.0	<0.001**
Abnormal cephalic presentations†	18.5	8.0	<0.001**
Breech presentations	12.0	5.4	0.011*
Transverse positions	1.1	0.5	0.994
Apgar 5 score			
Apgar score 9–10	83.0	93.2	0.001**
Apgar score 7–8	13.8	5.7	0.002**
Apgar score <5	3.3	1.1	0.074
Umbilical artery pH-value			
>7.20	65.4	88.2	<0.001**
7.10–7.20	23.2	10.0	0.004**
<7.10	11.5	1.8	<0.001**
Postnatal intensive care unit surveillance	28.7	9.3	<0.001**
	Craniosynostoses 2000–2007 (n = 51)‡	Birth registry Tyrol 2000–2007 (n = 57,317)	P-value
Vaginal mode of delivery	58.8	67.7	0.217
Vacuum extraction	11.8	7.9	0.445
Forceps delivery	0.0	0.0	0.929
Vaginal breech deliveries	3.9	0.2	<0.001**
Cesarean delivery	25.6	24.0	0.931
Episiotomy	44.7	39.9	0.654
Perineal tears	25.6	29.0	0.592
Labial/clitoridal lacerations	2.6	2.6	0.911
Vaginal tears	7.7	1.7	<0.001**

**P < 0.01; *0.01 < P < 0.05.

†Abnormal cephalic presentations include occiput posterior position, deflexion position, high stage longitudinal position, asynclitism and rare abnormal cephalic presentations [18].

‡Non-significant linear trend test within the birth registry data (2000–2007).

The secondary cesarean delivery rate including emergency procedures in the population with craniosynostosis was 16.7% and higher than in the general population [OR 1.9 (95% CI 0.84–4.39)]. Regarding the neonatal outcome of the different delivery modes in children with craniosynostosis the rate of children presenting with an Apgar 1 score of <5 was significantly higher in the group of secondary cesarean deliveries (50.0%) compared to vaginal deliveries (4.1%; P < 0.001) and primary cesarean deliveries (0.0%; P < 0.05). By contrast, the rate of newborns showing a pH-value of more than 7.30 was significantly higher in primary cesarean deliveries (80.0%) compared to vaginal deliveries (27.5%; P < 0.05) and slightly higher compared to secondary cesarean deliveries (40.0%; P = 0.52).

Children diagnosed with premature craniosynostosis showed a significantly higher rate of *fetal malpresentations* [31.52%; P < 0.001; OR 2.38 (95% CI 1.53–3.70); Tables 1 and 2], whereas the rate in the Tyrolean population was only 14.0%. This was mainly due to the remarkably high percentage of abnormal cephalic presentations [18.5%; P < 0.001; OR 3.42 (95% CI 2.03–5.76)], especially the high frequency of deflexions (12.0%). The rate of breech presentations (12.0%) was also significantly increased [P = 0.011; OR 2.39 (95% CI 1.27–4.49)]. The rate of transverse positions (1.1%) was slightly increased, however, the disparity was not significant (P = 0.99; Tables 1 and 2). In the group of premature craniosynostoses only 68.5% of all children presented with an ordinary occiput anterior position. Analysis of the delivery of fetuses with malpresentations reveals that abnormal fetal presentations were associated with several perinatal complications. Specifically, the strong coincidence of vaginally delivered breech presentations with dystocia (of 50.0%) is exceptional.

In our series *various types of premature craniosynostoses* have been observed, characterized by different suture involvement and resulting in specific skull deformities (Figure 1). The entity of the plagiocephaly is the malformation showing the highest rate of fetal malpresentations. More than

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Table 2 Odds ratio (OR) for development of malpresentations at birth in fetuses with craniosynostosis.

Characteristics	OR	95% CI	P-value
Fetal malpresentation	2.38	1.53–3.70	$<0.001^{**}$
Abnormal cephalic presentation[†]	3.42	2.03–5.76	$<0.001^{**}$
Breech presentation	2.39	1.27–4.49	0.011^{*}
Transverse position	2.12	0.29–15.29	0.994

$^{**}P<0.01$; $^{*}0.01<P<0.05$.

[†]Abnormal cephalic presentations include occiput posterior position, deflexion position, high stage longitudinal position, asynclitism and rare abnormal cephalic presentations [18].

CI = confidence interval.

half of all children diagnosed with plagiocephaly (53.9%) had an abnormal presentation, especially breech. Additionally, children with scaphocephaly also showed a high frequency of malpresentations (34.0%). In contrast, children presenting with a trigonocephaly never showed any abnormal fetal presentations (Table 3).

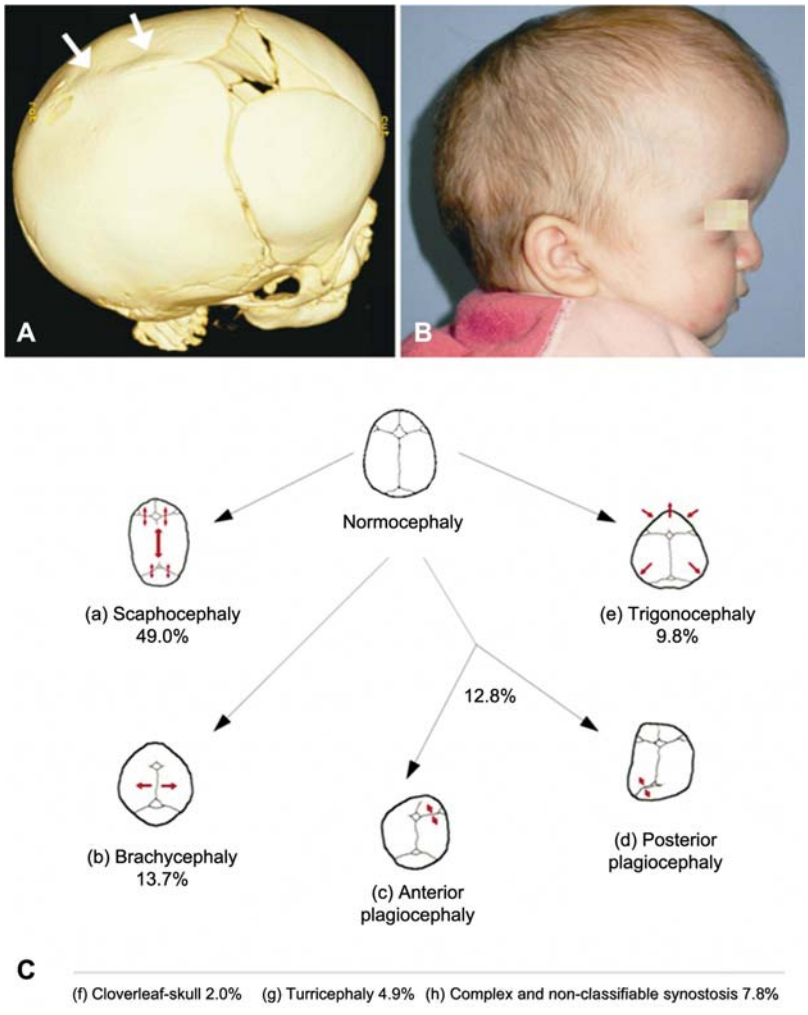

Figure 1 (A) Premature fusion of the sagittal suture in a three-dimensional-CT reconstruction in a child with scaphocephaly. Arrows indicate the fused sagittal suture. (B) Clinical picture of scaphocephaly. (C) Fraction of different craniosynostosis entities in the study population (n = 102) and affected cranial sutures resulting in specific skull deformities in superior view: (a) Scaphocephaly: premature synostosis of sagittal suture; (b) brachycephaly: premature synostosis of coronal suture; (c) synostotic anterior plagiocephaly: unilateral premature synostosis of coronal suture; (d) synostotic posterior plagiocephaly: unilateral premature synostosis of lambdoid suture and (e) trigonocephaly: premature synostosis of metopic suture.

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Table 3 Rates of fetal presentations in four different entities of craniosynostoses in (%).

Fetal presentation	Scaphocephaly (n = 50)	Plagiocephaly (n = 13)	Trigonocephaly (n = 10)	Brachycephaly (n = 11)
Abnormal cephalic presentation[†]	20.0	30.8	–	9.1
Breech presentation	12.0	23.1	–	9.1
Transverse position	2.0	–	–	–
Occiput anterior position	66.0	46.1	100.0	81.8

[†]Abnormal cephalic presentations include occiput posterior position, deflexion position, high stage longitudinal position, asynclitism and rare abnormal cephalic presentations [18].

In 44.7% of all births in the craniosynostosis group an *episiotomy* was performed. The difference was not statistically significant compared to the rate in Tyrol (39.9%; $P = 0.65$). Also the number of mothers suffering from various degrees of *perineal tears* (25.6%) and *labial* or *clitoridal lacerations* (2.6%) was not significantly increased (all $P > 0.05$). Only the frequency of *vaginal tears* (7.7%) was significantly higher in mothers of children with craniosynostosis compared to the rate in the Tyrolean population (1.7%; $P < 0.001$).

In 6.9% of all births in the craniosynostosis group a *cephalhematoma* was reported. However, in 71.4% of these cases the cephalhematoma was likely due to the use of vacuum extraction as the mode of delivery for these children. Other reported perinatal complications include failure to progress (5.9%) and dystocia (3.9%).

In order to compare the vitality of the neonates the *Apgar score* after 5 min was subdivided into five sections. As few as 83.0% of neonates of the craniosynostosis group presented with an optimal 5-min Apgar score between 9 and 10. This percentage is significantly lower (10.2%) than that of the general population (93.2%; $P = 0.001$). On the other hand, the rate of infants showing a 5-min Apgar score of 7–8 (13.8%) was significantly higher in the craniosynostosis group (5.7%; $P = 0.002$; Table 1). This implies that in about 10% of all children diagnosed with craniosynostosis the Apgar score was found to be at least one category lower than in the comparison population ($P < 0.001$). However, the extremely low 5-min Apgar scores (< 5), were not different between craniosynostoses and the comparison population ($P = 0.074$; Table 1).

Of the infants with craniosynostosis, 65.4% presented with an *umbilical artery* pH-value of > 7.20, which is lower than the rate reported in the general population (88.2%; $P < 0.001$). The frequency of infants having a pH-value between 7.10 and 7.20 was significantly higher in infants with craniosynostosis ($P = 0.004$) and, importantly, the rate of newborns presenting with a pH-value < 7.10 (11.5%) was higher compared to the general population (1.8%; $P < 0.001$; Table 1).

In 22.1% of all craniosynostosis patients different *fetal* or *neonatal abnormalities* were documented in the Austrian Mother-Child Pass. Besides the presence of a distinct cyanosis in 5.3% of newborns with craniosynostosis, an especially high rate of breathing complications (19.0%) was reported in the sample, with exceptionally high frequencies in those with brachycephaly (30.0%) and turricephaly (50.0%). Of those children presenting with complications of breathing, 88.2% also required neonatal intensive care treatment. Other documented characteristics include fetal hypoxia (8.2%) and neonatal icterus (3.2%).

One tenth of all neonates in the craniosynostosis group needed *primary resuscitation*. It is important to note that ''primary resuscitation'' according to the Austrian Mother-Child Pass also includes oxygen bag ventilation. However, 80.0% of these resuscitated infants also required *neonatal intensive care surveillance*. In total, 28.7% of craniosynostosis patients required postnatal intensive care treatment and monitoring, which is significantly higher than the rate of the general population [9.3%; $P < 0.001$; OR 3.95 (95% CI 2.52–6.17); Table 1]. This postnatal intensive care treatment was clearly associated with breech presentations as 58.3% of these children required intensive care after birth. Another association was seen in children diagnosed with brachycephaly as 64.7% of all children without a malpresentation but requiring postnatal intensive care treatment showed a brachycephalic malformation of the skull.

In addition to the breathing complications mentioned above, other *postpartum abnormalities* were documented in 20.8% of children with premature craniosynostosis. This includes complications such as fever (5.9%), respiratory infections (3.9%) and electrolyte disorders (2.9%). In 8.3% of patients the underlying anatomical malformation of the child led to a disturbance and cessation of maternal breastfeeding.

Only in 11 children (10.8%) of the study population an abnormal shape of the skull or even the definite *antenatal sonographic diagnosis* was documented in the Mother-Child Pass. The antenatal detection was usually based on the abnormal shape of the fetal skull in the ultrasound screening examination of the third trimester between the 28th and 32nd week of gestation (mean = 31; SD = 6.3). A common mode of delivery in this sample of prenatally diagnosed cases was cesarean delivery, which was performed in 45.5% of all births compared to 22.6% in postnatally diagnosed patients. The overall outcome of these cesarean deliveries of prenatally diagnosed children showed no major complications, whereas the vaginal deliveries were associated with several perinatal complications, such as cephalhematomas (16.7%), dystocia (16.7%) and perineal ruptures (33.3%).

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Discussion

The low rate of antenatal diagnosis (of about 11%) in conjunction with the exceptionally high rate of perinatal complications reveals a tremendous need for the awareness of premature craniosynostoses among prenatal and maternal-fetal-medicine specialists. This is supported by the fact that the reported complications in craniosynostosis patients are potentially reduced or even prevented by adapted perinatal management.

In the group of craniosynostoses, the rate of children presenting with an ordinary occiput anterior position was significantly lower than the rate seen in the general Tyrolean comparison population, whereas the rate of infants presenting with a malpresentation was significantly higher. However, it is important to note that these types of complications are not limited to children with craniosynostosis. In children with other malformations of the skull a higher rate of fetal malpresentations, in particular breech presentations, has also been observed. Interestingly, the association between breech presentation and hydrocephaly or anencephaly is commonly known [10, 12, 15, 22, 26]. According to Lawson [12], one-third of all fetuses suffering from hydrocephaly presents with a breech presentation at the onset of labor.

The most prevalent form of malpresentation in children with premature craniosynostosis in this study was the deflexion position. However, only in two cases it was possible to identify the actual subform of the malpresentation with one child presenting with a face presentation, and the other child with a brow presentation. In both of these abnormal fetal presentations, that occur in $<1\%$ of all births, cephalopelvic disproportion and an abnormal shape of the fetal skull have already been discussed as etiologic factors [13].

It is commonly known that malpresentation, such as breech, may be associated with several perinatal complications [7, 8, 20]. In the population of children with craniosynostosis analyzed in the current study, a strong relationship between abnormal fetal presentations and perinatal complications was seen. This relationship was most evident in children with breech presentations, which also underlines the obstetric relevance of these pathologies for affected children and their mothers. This is further supported by the fact that 58% of all children with breech presentation required postnatal intensive care treatment.

A high rate of children with postnatal cephalhematomas was seen in this study, which is certainly due to the comparatively high frequency of children being delivered by vacuum extraction in this sample (71.4%). This high rate of cephalhematomas, the relatively high rate of maternal perineal lacerations, and the significantly increased rate of vaginal tears also suggest that one major perinatal complication seen in children with premature craniosynostosis might be a hindered passage of the fetal head through the birth canal.

Our results indicate that in about 10% of children with craniosynostosis the Apgar score as well as the pH-value were found to be at least one category lower than in the general Tyrolean population. These results suggest that both the vitality and the oxygenation of about 10% of children with craniosynostosis were significantly reduced. Moreover, the rate of newborns requiring immediate neonatal intensive care was significantly increased in comparison with the general postnatal hospitalization rate in the Tyrolean population.

A possible explanation for these findings can be found in the high rate of postnatal breathing complications documented in 19% of all children with craniosynostosis. This is further supported by the additional 5% of children in which cyanosis was observed. Several other studies have already reported about the common need of airway management and tracheotomy in craniofacial anomalies in the first month of life [2, 21, 24]. However, a closer investigation shows that in this sample postnatal breathing complications as well as postnatal intensive care are closely associated with brachycephaly and turricephaly. This could be explained by the fact that in brachycephaly the sutures of the viscerocranium can also be affected [4, 23], which may lead to severe airway stenosis following maxillary hypoplasia.

There were no differences concerning the mode of delivery in children with craniosynostosis and in the general population. Also the overall cesarean delivery rate was not different. However, interesting details can be seen when the rates of primary and secondary cesarean deliveries is examined. The rate of secondary emergency cesarean deliveries was about 17% higher in children with craniosynostosis than in the general Tyrolean population. This high rate of secondary cesarean deliveries and the pattern of other perinatal complications in craniosynostosis patients indicate that the underlying craniofacial malformation leads to difficult vaginal deliveries in these children. This could only be prevented by a definite antenatal sonographic detection of craniosynostoses and a thorough consideration by physicians of performing primary cesarean delivery. The considerably low rate of primary cesarean deliveries is particularly intriguing when considering the exceedingly high rate of fetal malpresentations and its associated complications, as well as the significantly increased rate of vaginal breech deliveries. In addition, the reduced postnatal vitality and oxygenation further support the need for avoiding conventional vaginal deliveries as this might also improve outcomes by reducing perinatal stress to the fetus.

This issue becomes even more interesting when considering the advancements in prenatal diagnosis. According to Miller [17] and Delahaye [5] and their co-workers the definite antenatal ultrasound diagnosis of craniosynostosis is feasible for most entities from around the 2nd trimester of gestation. Results from the current study also support this conclusion, as suspicious prenatal sonographic findings were documented in about 11% of all craniosynostosis patients. Interestingly, the cesarean delivery rate in these patients was higher, whereas the perinatal complication rate in the surgically delivered children was lower.

Based on these findings we suggest that in fetuses with a sonographic suspicion of craniosynostosis and further obstetrically unfavorable criteria, an indicated cesarean delivery should be decided more generously in order to avoid haz-

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ardous operative vaginal deliveries as well as to improve the postnatal outcome of affected newborns by avoiding major perinatal complications.

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