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T. C. Moore · D. L. Collins · V. Catanzarite
E. I. Hatch, Jr.

Pre-term and particularly pre-labor cesarean section to avoid complications of gastroschisis

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Abstract The marked advantages and merit of pre-term and particularly pre-labor (PTPL) cesarean section (C-section) in the avoidance, and indeed, virtual elimination of severely disabling gastroschisis (GS) complications in infants diagnosed prior to birth by ultrasound has unfortunately remained controversial in the 10 to 12 years since it was first reported and strongly recommended by numerous authors. During this period, GS has remained one of the four major causes of the short-gut syndrome (SGS) in infancy and childhood and a major cause of prolonged, costly, complicated, and hazardous neonatal intensive care unit stays with requirements for total parenteral nutrition (TPN). The most serious and frequent complications of GS in infants born without PTPL C-section are the occurrence of the ''peel'', which greatly enlarges and rigidifies the eviscerated gut, and of ''complicated GS'' (intestinal atresia/s, stenosis, necrosis, perforations) (CGS). The ''peel'' occurs in 100% of these cases and CGS in approximately 20%. ''Peel'' enlargement and rigidification of eviscerated intestine in the presence of a reduced peritoneal cavity causes great difficulty in covering the eviscerated, enlarged, and rigidified gut with abdominal wall, skin, a prosthesis, etc., and frequently produces gut ischemia from excessive pressure, which may lead to necrotizing enterocolitis (NEC) and SGS as well as prolonged hospital stays. The presence of a ''peel'' greatly complicates the hazards of dealing with cases of CGS, as resection and anastomosis are virtually impossible in the presence of a ''peel.'' The authors report personal experience with 77 cases of GS dating as far back as 1951; 44 of the infants were born after the onset of labor by vaginal or C-section delivery and all had some degree of ''peel'' formation. Of 320 cases from the literature (including some of the cases reported here), 61 (19.1%) involved CGS. Of the 33 cases born PT, and especially PL, there were no cases of ''peel'' and only 1 case of CGS (3.0%). This infant had a single atresia associated with a very small (1 cm) defect in the abdominal wall and no labor-induced ''peel,'' which was easily and successfully repaired by resection and anastomosis. The 6.4-fold reduction in the occurrence of CGS by PTPL C-section (3.0% vs 19.1%) was statistically significant by the chi-square test ($P < 0.05$), as was the 100% elimination of the disabling ''peel.'' If the single case of CGS associated with a very small defect and no labor or labor-associated ''peel'' is eliminated, the incidence of CGS in the remaining PTPL group of 32 cases falls to 0 (0% versus 19.1%, $P < 0.007$). PT and especially PL C-section may be expected to virtually eliminate ''peel'' formation and CGS and to remove GS as one of the four major causes of SGS. The findings of this report that PT labor prior to PT C-section may result in both ''peel'' formation and CGS further solidifies the role of labor in the production of both the ''peel'' and the equally disabling CGS. Failure to appreciate the central role of labor in GS complications has doubtless contributed to the persistent controversy concerning the value and importance of PTPL C-section for gastroschisis diagnosed in utero. The pediatric surgeon has an important responsibility with the obstetrician to monitor the possible occurrence of occult labor in the waning weeks of pregnancy and be prepared to do a prompt C-section if it occurs and there is adequate lung maturity. The achievement of ''peel''-

T.C. Moore (✉)
Division of Pediatric Surgery
Harbor-UCLA Medical Center
1000 West Carson Street,
Torrance, California 90509, USA

D.L. Collins
Department of Surgery,
Children's Hospital of San Diego,
San Diego, CA 92123, USA

V. Catanzarite
Sharp/Children's Prenatal Diagnostic Center,
8010 Frost St., Suite M,
San Diego, CA 92123, USA

E.I. Hatch
Department of Surgery,
Children's Hospital Medical Center,
4800 Sand Point Way NE,
Seattle, WA 98105, USA

and CGS-free gut would greatly facilitate the use of the new Bianchi technique of gut reduction without anesthesia. The combination of the use of epidural anesthesia for the elective PTPL C-section with the Bianchi approach would spare both mother and baby any untoward effects of general anesthesia and present the potential for massive reductions in hospital costs with minimal patient manipulation and disturbance. For infants born with labor-associated ''peel,'' re-evaluation of the suitability and effectiveness of surgical ''peel'' decortication from involved gut is strongly urged.

Key words Gastroschisis · ''Peel'' · Complicated gastroschisis · Short-gut syndrome · Pre-term labor

Introduction

Gastroschisis (GS) is a major malformation of the newborn involving a large and awesome-appearing evisceration at birth of intra-abdominal contents, largely small and large intestine, onto the anterior abdominal wall through a sacless midline defect to the right of the umbilicus. When birth occurs after the onset of labor (either vaginal or cesarean section [C-section]), the eviscerated structures tend to be discolored and often cyanotic (Fig. 1) and covered with a gelatinous-appearing matrix often described as a ''peel'' and often of a leathery consistency, greatly enlarging and rigidifying the eviscerated mass.

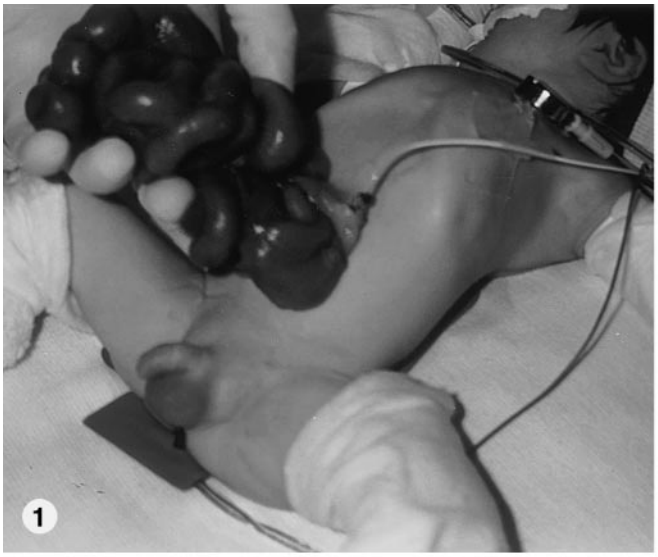
In 1953, Moore [24] reported two cases of GS and presented a classification of anterior abdominal wall eviscerations at birth that differentiated GS and omphalocele. At that time, there were less than a handful of cases of GS reported in the world literature. One of the cases in this report involved ''complicated GS'' (CGS) (multiple atresias imbedded in a huge, gelatinous matrix). The metabolizable nature of this large mass was demonstrated by its disappearance on skin-flap coverage within 18 days in association with the appearance of normal peristalsis. Reoperation at that time revealed normal-appearing small intestine (with the exception of the atresias), and a bypassing entero-enterostomy was successfully carried out without technical difficulty. The ''peel'' in the second case was considerably smaller, but still of significance, and on histologic study the ''peel'' was found to be extraserosal and of a granulomatous/edematous nature (Fig. 2). It was suggested by the author that the ''peel'' was most likely due to prolonged immersion of the eviscerated gut in amniotic fluid (almost certainly wrong from current data). An interesting observation was the association of the size and extent of the ''peel'' with the duration of labor. The infant with the huge ''peel'' had experienced 20 h of labor while the infant with a substantially lesser extent (and no CGS/multiple atresias) had experienced only 5 h.

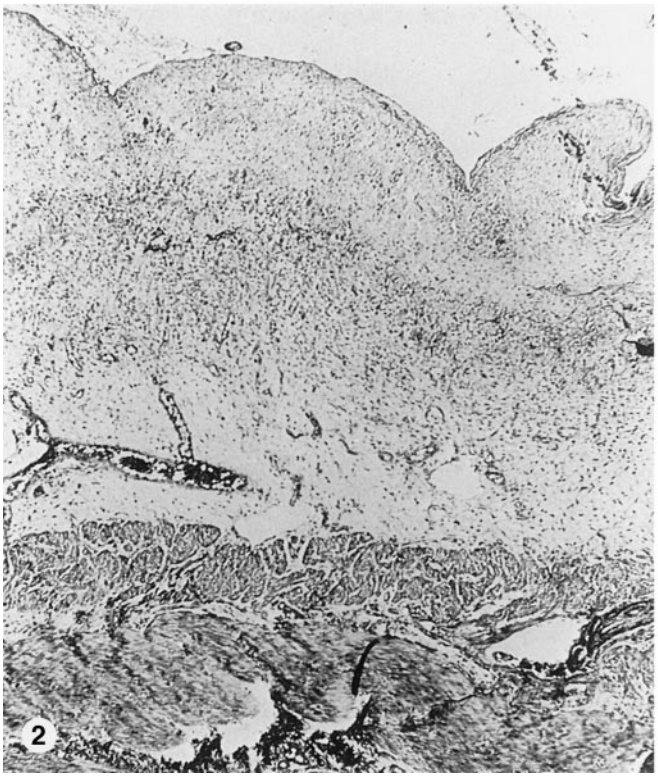
A 1963 report by Moore [17] introduced the word ''peel'' into the GS literature, as the author was able surgically to remove the thick, gelatinous matrix as a ''peel'' is removed in the decortication of a trapped lung. The great enlargement of the eviscerated mass was found to be due to entrapped sub-''peel'' edema, which was easily expressed during ''peel'' removal with the restoration of normal-appearing gut, which promptly resumed persistalsis on placement into the peritoneal cavity. A meconium stool was passed on the day following the operation. Good bowel sounds were heard after 3 days and oral feedings were started successfully 1 day later. This report introduced ''peel'' decortication as a therapeutic option in the surgical management of GS. Greatly improved electrocautery dissection since 1963 should considerably enhance the ease of this decortication in GS infants born after the onset of labor and be much more effective in dealing directly with the ''peel''-associated hazards of silos and the stuffing of the ''peel''-rigidified and enlarged gut mass forceably into a too-small peritoneal cavity.

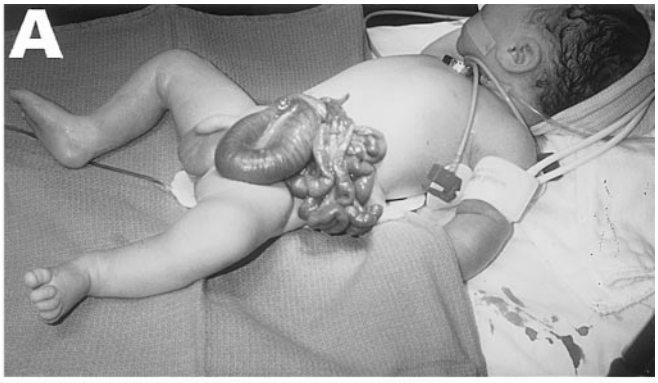
A 1977 report [18] provided additional evidence that GS and omphalocele (OMP) are quite distinct malformations and that GS is essentially free of additional malformations while OMP is associated with multiple, serious malformations including a number of OMP ''syndromes.'' This report also described an increased incidence of prematurity (60%) in GS as compared with OMP. Three additional reports in 1986 and 1987 [25–27] were based on an international survey that produced 490 cases for detailed study of the dynamics, including possible causes, of the GS ''explosion'' that was of almost epidemic proportions in terms of the numbers of new cases appearing in the literature during a limited period of time.

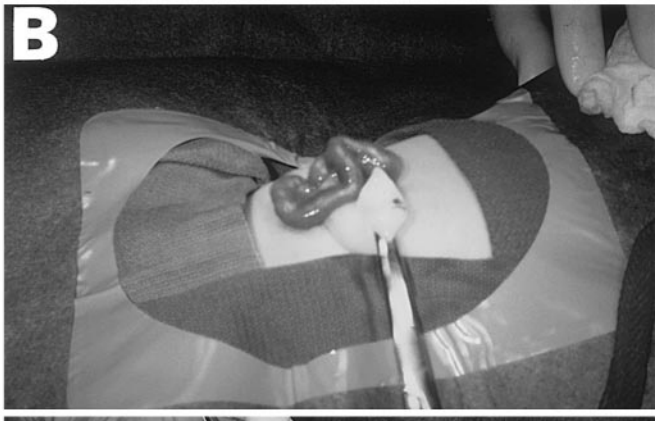
The development of ultrasound and its increasing use in obstetrics made possible not only the in-utero diagnosis of GS during pregnancy, but also new options and strategies for monitoring and delivery, including C-section. A major breakthrough in the management of GS occurred in 1986 with the publication (in the obstetric literature) by Lenke and Hatch [14] of clinical data clearly identifying the value of pre-term C-section in essentially eliminating GS complications in comparison with prior experience at the same institution with vaginal delivery after the onset of labor. In 1988, almost simultaneous reports by Hagberg et al. [12] and Moore [19] confirmed these findings. Hagberg et al. also eliminated prolonged immersion in amniotic fluid as a cause of ''peel'' formation: they made the important observation that infants whose GS was diagnosed in utero many weeks prior to term and managed by pre-term and prelabor (PTPL) C-section were consistently ''peel''-free. They considered the trauma of labor and delivery to be the cause of ''peel'' formation.

In a 1988 report by Moore [19], labor was suggested as the primary cause of ''peel'' formation. This judgement was further reinforced by a 1992 paper [21]. That report and the present one emphasized the primacy of labor in both ''peel'' formation and the occurrence of CGS, as infants with known GS in utero and born by









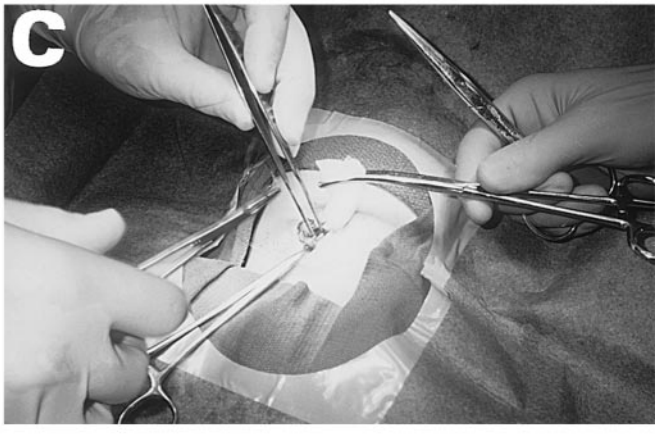


Fig. 1 "Peel"-covered, enlarged, discolored, rigidified gastroschisis mass in an infant born without pre-term and pre-labor C-section
Fig. 2 Photomicrograph of gut from a gastrochisis infant showing granulomatous/inflammatory and extra-serosal "peel"
Fig. 3 **A** "Peel"-free, non-enlarged, and normal-colored and -appearing intestine in an infant born by pre-term and pre-labor C-section; **B** easy placement of intestine into peritoneal cavity; **C** appearance of anterior abdominal wall at completion of placement

C-section after the onset of labor may still have both "peel" formation and CGS. A 1991 report [20] described the successful use of the "patch, drain, and wait" (PD&W) surgical approach in the management of a case of CGS with massive intestinal necrosis at birth. Decortication of the enlarged and rigidified mass in cases of lesser necrosis, multiple atresias, or both might be an additional option with added PD&W safeguards. A 1994 report by the same author [22] described the virtual elimination of CGS by PTPL C-section.

With the passage of 12 years since the Lenke and Hatch report and 10 years since those of Hagberg et al. and Moore and the persistence of controversy regarding the use and usefulness of the PT and particularly PL approach to GS diagnosed in utero, the authors have elected to "revisit" this important topic of PTPL C-section for the avoidance of GS complications.

Materials and methods

The focus of this report is on the value and usefulness of PT and particularly PL C-section in the avoidance/elimination of "peel" formation and CGS (intestinal atresia/s, stenosis, necrosis, perforations). The clinical data presented here represent the personal experience of three of the authors (TCM, EIH, DLC) plus that of the late Professor Sture Hagberg and associates at the University of Göteborg in Sweden. The first author (TCM) is indebted to Professor Hagberg for sharing the pre-publication data of his important work on this topic, including a color photograph showing the "peel"- and CGS-free appearance of a newborn with GS managed by PTPL C-section.

Results

Of 77 cases of GS from this combined experience, 44 were born after the onset of labor by either vaginal delivery or C-section and all 44 had "peel" formation (Table 1). None of the 33 infants with GS born by PTPL C-section were found to have "peel" formation (Table 1). Our chief statistician considered this difference highly significant. From a total of 320 cases of GS managed without PTPL C-section collected from major published series (including 12 cases from the present experience) 61 (19.1%) were reported to have CGS (Table 2). Atresia was the most frequent of these complications, occurring in 29 cases (9.1%), followed by necrosis/gangrene with 16 cases (5.0%) and perforation and stenosis with 5 cases each (1.6%).

Of the 33 cases of GS in this experience managed by PTPL C-section, CGS occurred in only 1 (3.0%) (Table 3), and involved a single atresia in an infant with a very small GS defect (approx. 1 cm). The 6.4-fold reduction in the occurrence of CGS associated with PTPL C-section as compared with delivery after the onset of labor (3.0% vs 19.1%) is statistically significant by the chi-square test [$P < 0.05$]. If the non-labor and non-"peel"-associated atresia with a small defect as a fairly certain cause of the atresia is eliminated from the PTPL patients with CGS, in the 32 remaining cases the incidence of CGS falls to 0% as compared with 61 of 320 others (19.1%), an even more statistically significant difference [$P < 0.007$].

Table 1 Incidence of "peel" formation in infants with gastroschisis born with and without pre-term and pre-labor C-section

	Period	No. of cases	"Peel"	% "Peel"
Without				
Lenke/Hatch [14]	1982–1984	17	17[a]	100[a]
Moore	1951–1987	17	17	100
	1996–1997	2	2	100
Moore [21]	1987–1995	8	8	100
	Total	44	44	100
With				
Moore [19, 21]	1987–1997	6	0	0
Collins	1992–1997	13	0	0
Lenke/Hatch [14]	1982–1984	7	0	0
Hagberg [12]	1984–1988	7	0	0
	Total	33	0	0

[a] All 17 infants were delivered vaginally after the onset of labor with extensive "peel"-type gut damage. There were 3 deaths, 2 cases of "complicated gastroschisis" (intestinal atresia, stenosis, necrosis, perforation), and primary closure was not possible in 6 cases. In 2 cases, bowel infarction (necrotizing enterocolitis) resulted in 1 death 25 days after primary closure and 1 case of total bowel resection, 4 months of hospitalization, and indefinite total parenteral nutrition. Appendectomy was carried out in only 2 infants, suggesting that bowel damage was particularly severe and extensive in this group

Photographs (TCM) of two successive newborns with GS managed by PTPL C-section in San Diego are presented in Figs. 3 and 4, and show key stages in the easy and rapid replacement of the "peel"- and CGS-free intestine into the peritoneal cavity. The umbilicus was preserved in both cases. The operating time was about 7 to 8 min in each case. In a recent 6-month period, one of us (TCM) has had personal experience with 5 cases of GS, 3 were managed successfully by PTPL C-section with no "peel," no CGS, and easy replacement of the "peel"-free and pliable gut into the peritoneal cavity without defect enlargement and with preservation of the umbilicus. Two were born prematurely at 33 and 34 weeks of gestation by C-section and after 2 weeks and 8 days of premature labor (one with ruptured membranes). Both had extensive "peel" formation [3+ and 4+] and one had CGS with a large area of necrosis (Fig. 5). Both have had greatly prolonged hospitalizations with total parenteral nutrition (TPN) and multiple complications. These last 2 cases provide information relative to the importance of labor in the generation and extent of both "peel" formation and CGS. They also suggest the importance of careful monitoring of known GS cases in utero for the occurrence of premature labor and its prompt and emergent management.

In a 1986 report [14], 17 infants with GS were delivered by the vaginal route after the onset of labor and during the same period of time as 7 PT C-section infants. The 7 PT C-section infants were both "peel"- and CGS-free. All had easy primary closure and limited, complication-free periods of hospitalization. In the simultaneous group of 17 GS infants delivered vaginally after the onset of labor, there were 3 deaths and 2 cases of CGS [1 perforation and 1 necrosis]. Attempted primary closure was not possible in 6 of the 17. In 2 patients in whom primary closure was carried out, bowel infarction necrotizing enterocolitis (NEC) resulted in 1 death 25 days after primary closure in 1 and total bowel resection was required in the other with 4 months of hospitalization up to the time of the report and a future of long-term TPN. Appendectomy was carried out in only 2 of the 17 infants born by the vaginal route and after the onset of labor, suggesting to the authors that bowel damage in this group was more severe and more extensive.

An intriguing observation by one of us (VC) was the presence of biochemical lung maturity at the time of initial amniocentesis in all cases of GS diagnosed in utero compared with an incidence of only 30% in other

Table 2 Occurrence of midgut necrosis/gangrene, perforation/s, atresia/s, and stenosis in reported cases of gastroschisis not managed by pre-term and pre-labor C-section

Author	Year	No. of cases	Necrosis/ gangrene	Perforation	Atresia	Stenosis	More than one	Total one or more	% one or more
Moore [24]	1953	2			1			1	50
Lewis [15]	1973	31	6		2			8	26
Hollabaugh [13]	1973	47		1	6			7	15
Amoury [1]	1977	46	1				5	6	13
Pokorny [29]	1981	22	1		5			6	27
Luck [16]	1985	106	3	3	7	3		16	15
Tibboel [31]	1986	6			1	2		3	50
Lenke [14]	1986	17	1	1				2	12
Bond [4]	1988	11	2		2			4	36
Gornall [10]	1989	22			4		1	5	23
Moore [21]	1992	8	1		1			2	25
Moore	1997	2	1					2	50
Total		320	16	5	29	5	6	61	19.1

cases at a comparable period (34 weeks' gestation) not associated with GS.

Discussion

In the 1953 report by Moore [24], the correlation of both the degree of "peel" and the presence of CGS and its degree of severity (multiple atresias) with the length of labor were recorded. As these important complications of GS appear increasingly to be related not only to labor, but to its duration and intensity as well, it is of increasing importance that the last trimester of pregnancy be monitored with increasing care by the pediatric surgeon as well as the obstetrician despite the inconvenience of their often being located in different areas, buildings, or units. The accumulation of more precise and accurate data correlating the degree of "peel" and the duration of labor in antenatally-diagnosed cases of GS is of considerable importance in future studies.

The findings of this report clearly demonstrate the marked superiority of PT and particularly PL C-section in the management of GS diagnosed in utero. This superiority is primarily related to the essential elimination of the disabling, gut-enlarging, and rigidifying "peel" (likely labor-induced with an extent and degree influenced by the duration and strength of contractions) and the marked reduction in the occurrence of CGS (intestinal atresia/s, stenosis, necrosis, perforation).

The term CGS was first and aptly used in an important 1990 report from Columbus, Ohio, by Caniano et al. [6], who reported that 10 of their 80 cases of GS seen within a 7-year period (1979–1986) (12.5%) were "complicated." Three of the 10 infants with CGS had intrauterine volvulus and intestinal atresia with a residual jejunoileum of 10, 17, and 50 cm; 3 had ileal atresia, 2 had jejunal atresia with adequate length, and 2 had atresia with 45 and 55 cm of jejunoileum. Two of these infants died (20%). Four infants with adequate intestinal length and 4 of the 5 with short-bowel syndrome (SBS) were long-term survivors. The average period of hospitalization for the 4 infants with adequate intestinal length was 65 days; 3 had serious postoperative complications involving sepsis. The 4 infants with SBS all experienced multiple and serious complications and had prolonged initial hospitalizations ranging from 8 months to 3 years. The fate of the 70 cases of "uncomplicated" GS was hardly benign: about half each were treated by construction of a prosthetic silo (36 cases, 51%) or primary closure (34 cases, 49%). The average length of hospitalization for the silo group was 56 days (range 26–177) and the average cost $39,900. For the primary

Table 3 Occurrence of midgut necrosis/gangrene, perforation/s, atresia/s, and stenosis in reported cases of gastroschisis managed by preterm and pre-labor C-section[a]

Author	Year	No. of cases	Necrosis/ gangrene	Perforation	Atresia	Stenosis	More than one	Total one or more	% one or more
Lenke	1986 [14]	7	0	0	0	0	0	0	0
Moore	1988 [19] 1992 [21]	4	0	0	0	0	0	0	0
Hagberg	1988 [12]	7	0	0	0	0	0	0	0
Moore	1997	2	0	0	0	0	0	0	0
Collins	1997	13	0	0	1	0	0	1	7.7
Total		33	0	0	1	0	0	1	3.0

[a] All cases diagnosed by antenatal ultrasound

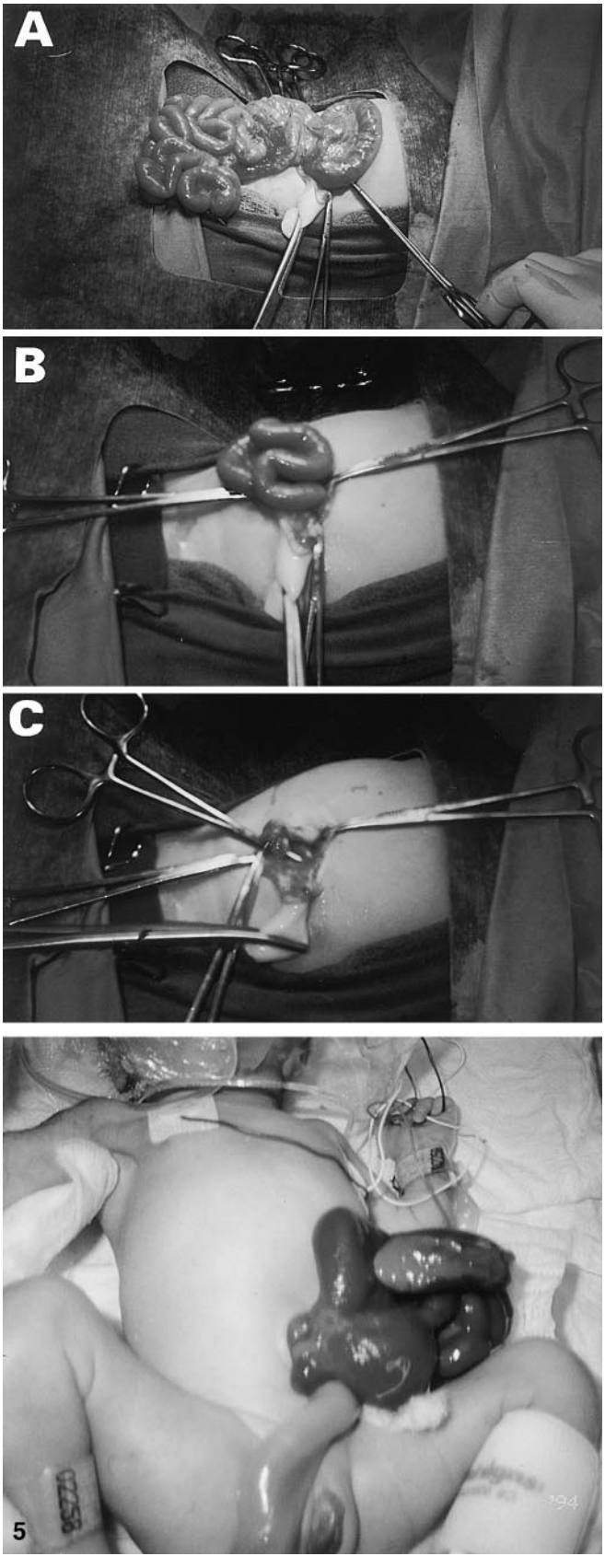


Fig. 4 **A** Infant with gastroschisis born by pre-term and pre-labor C-section with "peel"-free, normal-appearing intestine; **B** easy placement of intestine into the peritoneal cavity; **C** at completion of placement
Fig. 5 Gastroschisis infant born at 33 weeks' gestation after 8 days of labor and ruptured membranes showing marked "peel" formation and extensive necrosis and cyanotic discoloration of a loop of intestine ("complicated gastroschisis")

closure group, the average hospital stay was 47 days (range 14–132) and the average cost $32,800. They gave no cost figures for the 10 cases of CGS. These figures of 12 to 19 years ago translated into today's neonatal intensive care/TPN dollar figures would be truly astronomical!

In an earlier report from this Columbus group in 1973, Hollabaugh and Boles [13] described an experience with 47 infants with GS, 7 of whom (15%) had CGS (6 atresias and 1 perforation). The surgical management of these infants was greatly compromised by the presence of the "peel," and all 7 died. In a 1977 report from Kansas City by Amoury et al. [1], CGS occurred in 6 of 46 newborn GS infants (13%) with a mortality of 67% (best result up to that time!). In this experience intestinal atresia, necrosis/gangrene, and perforation occurred in 2 infants, single atresias and marginal viability or necrosis in 2, multiple atresias and gangrene in 1, and necrosis only in 1 with atresia developing post-anastomosis 1 cm proximal to a still-patent anastomosis. The authors interpreted these findings as suggesting that these "complications" were of rather late onset in the pregnancies and were still ongoing in some cases even at the time of birth. The findings could also rather easily be interpreted as "labor-induced."

Stuffing "peel"-enlarged and rigidified gut into too small a peritoneal cavity carries the hazard of inducing gut ischemia and NEC. Oldham et al. [28] of Ann Arbor reported a surprisingly high incidence of NEC following repair of GS. The occurrence of NEC in two consecutive infants after GS repair prompted a retrospective review of their experience in 54 newborns in an 11-year period. Ten of the 54 infants (18.5%) developed NEC, with 21 distinct episodes of NEC occurring in up to 3 episodes per patient (mean 2.1). In 1988, Ein et al. [7] from Toronto, Winnipeg, and Gainesville reported the occurrence of four cases of ischemic bowel after primary closure of GS in a 3-year period. All four were re-explored and necrotic bowel was found. SBS resulted in 2 cases.

Both "peel"-associated induced NEC and CGS may contribute to the development of the SBS in infancy and childhood. In recent reports dealing with causes of SBS in this age group, GS consistently appears as one of the four major ones [5, 8, 9, 11]. In a recent report from the University of Pittsburgh by Reyes et al. [30], GS was cited as the most frequent single indication for small-bowel transplantation in their experience with 55 such cases in children. From the findings of these reports as well as our experience, PT and particularly PL C-section should virtually eliminate GS as one of the four major causes of the SBS in infancy and childhood.

An important paper regarding the nature of the GS "peel" was published in 1988 by Amoury et al. [2]. They described the histologic appearance of the "peel" in ten cases of gastroschisis and found it to be extra-serosal and composed of fibrin and collagen based on special stains, quite similar in appearance to that pictured in Fig. 2. A correlation of these findings with the duration of labor would be of interest in newborns born after the onset of labor and with the histologic appearance of operatively-decorticated "peel."

The ease and speed of placement of the "peel"- and CGS-free gut into the peritoneal cavity in less than 10 min following PT and particularly PL C-section described in the Results section (Figs. 3 and 4) and the remarkable reduction in cut-and-sew operating open challenging opportunities for major reductions in costs and periods of hospitalization. A new technique of GS reduction without anesthesia reported in 12 cases by Bianchi and Dickson [3] of Manchester at the 1997 meeting of the British Association of Paediatric Surgeons in Istanbul would be especially applicable in GS in patients born by elective PTPL C-section. The use of epidural anesthesia for these C-sections and the Bianchi approach to the placement of eviscerated gut (now "peel"- and CGS-free) into the peritoneal cavity may spare both mother and baby any untoward effects of general anesthesia. With this approach, the potential exists for massive reductions in hospital care and costs, minimal patient manipulation and disturbance (including earlier "bonding"), as well as the possibility of it being done on an outpatient basis.

The intriguing observation cited at the end of the Results section regarding lung maturity identified biochemically at 34 weeks' gestation in all GS pregnancies compared with only approximately 30% of non-GS pregnancies suggests a number of additional studies such as monitoring amniotic-fluid pressures and growth factors, including lung-growth factors. These findings may suggest new strategies and options for use in experimental and clinical congenital diaphragmatic hernia, where tracheal-fluid pressures and lung growth and likely lung-growth factors are of great importance. It has recently been suggested by Moore [23] that it might be simpler and safer in congenital or experimental in-utero-acquired diaphragmatic hernia to increase amniotic-fluid and intratracheal lung-fluid pressures by infusing crysatalloids or colloids directly into the amniotic sac while monitoring amniotic-fluid pressures and biochemistry, including growth factors.

Addendum Since this paper was submitted for publication, an important report on this topic has been given at the Annual Meeting of the American Pediatric Surgical Association in Hilton Head, South Carolina (10–13 May 1998) by Moir and associates of the Mayo Clinic: A prospective trial of elective pre-term delivery for fetal gastroschisis [Abstract 32]. The key observations of this abstract report are shown in Table 4.

Table 4 Elective pre-term delivery for gastroschisis [Moir et al.]

	Pre-term (n = 16)	Non-pre-term (n = 16)
Significant differences		
Age at delivery (weeks)	34.2	37.7
Serious bowel compromise	0%	70%
Need for silo	0%	77%
Wound complications	0%	23%
Time to full enteral feedings (days)	19.1	35.1
Duration of TPN (days)	18.7	34.7
Time to hospital discharge (days)	22.7	37.7
No significant differences		
Birth weight (g)	2,308	2,409
Ventilator dependence (days)	1.3	1.4
Prematurity disabilities	0	0

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