Circumcision: A minor procedure?

Paediatric Death Review Committee: Office of the Chief Coroner of Ontario

A male infant at 37 weeks' gestation was born as a spontaneous vertex weighing 3.9 kg, with Apgar scores of seven at 1 min and nine at 5 min. Prenatally mild bilateral dilation of renal pelvises and ureters was noted on ultrasound. A urology consultation at 36 weeks' gestation suggested a possible normal variant and recommended a kidney ultrasound at three to five days postdelivery, with a voiding cystourethrogram to be performed if the dilation persisted.

The baby was bottlefed and was reported to be doing well when he was circumcised using a PlastiBell ring (Hollister, USA) at 10:00 on the seventh day of life (Figure 1). Local anaesthetic was not used. Some slight oozing was noted during the procedure but it was not a problem at discharge.

Five hours later, the parents returned to their family doctor with the infant, reporting that he was very irritable and had blue discolouration below the umbilicus when he cried. The doctor confirmed this observation and noted that the penis was slightly swollen. The abdomen was soft and nontender with bowel sounds noted. He was seen in-hospital shortly after by a paediatrician, who again noted the blue discolouration below the umbilicus, especially with crying, and some slight swelling of the penis. He had several loose stools, and it could not be determined with certainty whether he had voided. His temperature was 37.8°C and his respiratory rate was 38 breaths/min. There was good capillary refill in the legs; they were warm, and peripheral pulses were palpated. His blood pressure was 80/65 mmHg in the arm and 120/75 mmHg in the leg, and his heart rate was 196 beats/min. The baby was subsequently discharged home.

The infant was brought to a different hospital 14 h after the circumcision. He was now noted to be extremely irritable, with marked swelling of the penis and with bruising, swelling and cyanosis of the scrotum and perineum. He had grunting respirations and was cyanotic below the umbilicus. His temperature was 38.8°C, heart rate 190 beats/min, respiratory rate 24 breaths/min, saturation 98% in room air and blood pressure 104/34 mmHg. A blood culture was obtained, which subsequently grew *Escherichia coli* sensitive to ampicillin and gentamicin, which were started at this time. His white blood cell count was 2.2×10^9/L, with 35% bands. The immature to total neutrophil ratio was 1.3. His platelet count was 221×10^9/L and hemoglobin was 163 g/L. He was transferred to a tertiary care centre, where the bladder was identified as being distended to the level of the umbilicus. The PlastiBell ring was removed 16 h after the circumcision and a catheter was passed. The bladder was drained and the bluish coloration below the umbilicus subsided. Urine volume was not recorded. Over the next few hours, the infant went into septic shock with purpura fulminans, and went on to develop multiorgan failure and disseminated intravascular coagulopathy, with a partial thromboplastin time of 1.01 min, an international normalized ratio of 2.63 and a platelet count of 80×10^9/L. He died seven days after his circumcision from hypoxic-ischemic encephalopathy. The infant was not considered to have necrotizing fasciitis.

At autopsy, mild dilation of both kidney pelvises and ureters was noted. The bladder and urethra were normal.

Complications of meatal obstruction with the PlastiBell technique have been previously described in the literature (1,2). Necrotizing fasciitis as a complication of circumcision is rare, and all cases reported seem to be associated with the PlastiBell technique (2,3). The finding of cyanosis below the umbilicus after circumcision due to meatal obstruction caused by a misplaced PlastiBell ring resulting in bladder distension and obstruction of venous return has also recently been described (1). A review of circumcision complications suggest that these may occur more frequently than is conventionally believed (2,4).

The members of the Paediatric Death Review Committee of the Office of the Chief Coroner of Ontario were collectively aware of complications from their own institutions, including two children with necrosis of the glans, two infants requiring transfusion, one infant with a buried penis secondary to circumcision, numerous cases of retention of a PlastiBell ring, one infant with a slipped PlastiBell ring causing a penile tourniquet, and one infant with meatal obstruction due to a misplaced PlastiBell ring (Figures 2A and 2B). None of these complications were reported in the medical literature and are therefore not available in a retrospective literature review. It is concerning that none of the initial three physicians who saw this infant, including the physician who performed the procedure,

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Accepted for publication February 13, 2007

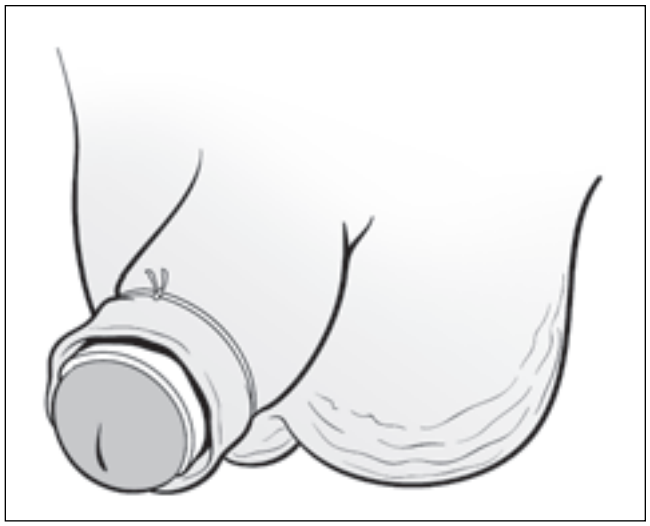

Figure 1) *Correctly positioned PlastiBell ring (Hollister, USA)*

identified this problem as a meatal obstruction, although they were all concerned about a possible link with the recent circumcision.

In this infant, there was no description of the glans by the physician removing the PlastiBell ring. If the foreskin is pulled too tight, then there will be considerable tension pulling the ring against the glans, thus compressing the urethra and making urination difficult or impossible. This is the mechanism described in the case report by Ly and Sankaran (1). We propose the mechanism shown in Figure 2B, given the rapid onset of symptoms, suggesting a complete obstruction. These could be differentiated clinically. In Figure 2A, the meatus would be visible in the middle of the ring and a catheter would be difficult or impossible to pass. In Figure 2B, the meatus would not be visible. The management of both complications would be the immediate removal of the PlastiBell ring. Perhaps a prospective surveillance study by the Canadian Paediatric Society of the complications of circumcision is warranted. Such a study would provide more accurate information for the ethical requirement of informed consent.

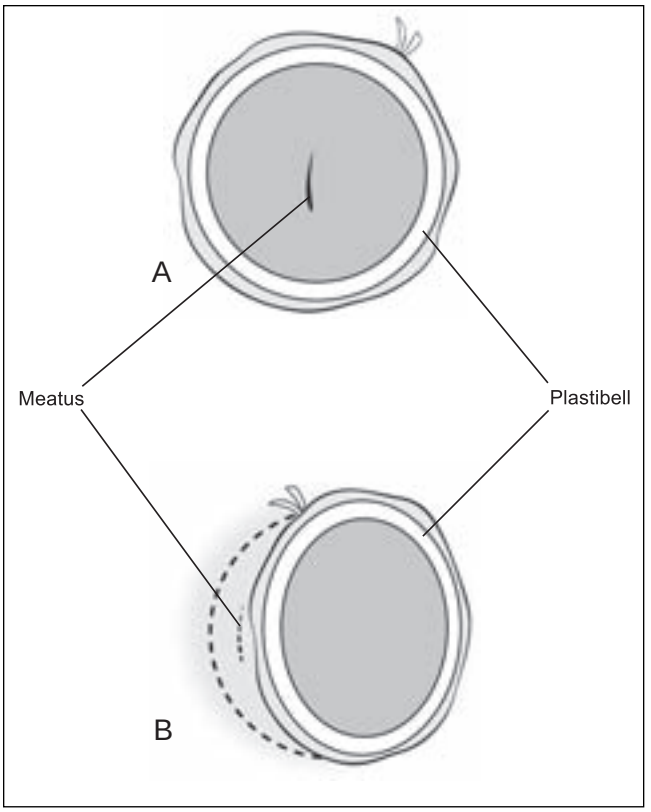

Figure 2) A *Slipped PlastiBell ring (Hollister, USA) causing tourniquet;* **B** *Slipped PlastiBell ring causing meatal obstruction*

ACKNOWLEDGEMENTS: The Paediatric Death Review Committee thanks the parents for their permission to report this case, and the medical graphics department at the Hospital for Sick Children for the illustrations.

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