CASE REPORTS

Penile Necrosis due to Priapism Developed After Circumcision in a Patient with Protein S Deficiency

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ABSTRACT

Introduction. Hypercoagulable state is a complex condition with an abnormal propensity for thrombosis. The consequences of it due to thrombosis of veins and arteries are the most common cause of sickness.
Aim. The present study is a report of a case describing penis necrosis after circumcision and evaluation of hypercoagulable state as a reason of it.
Methods. Nine-year-old boy referred from another hospital with the sequelae of the penile ischemia with discoloration of the penis after traditional circumcision.
Results. The ischemic event developed 3 weeks after circumcision. Priapism that was treated with needed glandulocavernous shunt initially developed following circumcision. All values of routine blood count and blood biochemical analysis were within normal limits. Further, hematologic studies revealed that there might be a hypercoagulable state as a result of lower protein S level.
Conclusion. Circumcision is a common procedure frequently performed in many communities around the world. Although it is seen as an easy procedure and performed even by unauthorized medical stuff, it is not complication-free. Severe penile necrosis after circumcision should be treated on an individual basis, necessitating different techniques. Hypercoagulable state should be concerned and evaluated in such a complication. In short, circumcision has to be done by specialists who can handle such serious complications. **Canter HI and Coskuner ER. Penile necrosis due to priapism developed after circumcision in a patient with protein s deficiency. J Sex Med 2011;8:3236–3240.**

Key Words. Circumcision; Necrosis; Protein S; Hypercoagulable State; Priapism

Introduction

Complications from traditional male circumcisions have been reported to be hemorrhage, infection, meatal stenosis, urethro-cutaneous fistula, shortening of the shaft skin, abnormal scarring, psychological grief caused by loss of the prepuce, and partial or total loss of prepuce [1].

In general, hypercoagulable states are associated with increased risk of surgical complications. Protein S deficiency in such a state in which beside recurrent venous thrombotic events, catastrophic neonatal purpura fulminans, and arterial thrombotic events associated skin necrosis have been reported [2–4]. Protein S acts as a cofactor for protein C, which is a vitamin K-dependent protein, like protein S, with serine proteinase action. With this proteinase action, these two proteins have a role in the inactivation of factors Va and VIIIa, and therefore they contribute to the coagulation activity.

The purpose of this article is to present a case of priapism and penis necrosis developed after traditional circumcision in a 9-year-old child with hypercoagulable state, which was characterized by protein S deficiency, diagnosed after the incident.

Method

An otherwise healthy 9-year-old boy, referred from another hospital with the sequelae of the

Figure 1 Appearance of the genital area of the patient at admission to the hospital.

penile ischemia with discoloration of the penis after traditional circumcision, was seen at our pediatric emergency department (Figure 1). His medical history revealed that the patient had been circumcised with dorsal slit technique under regional anesthesia with dorsal penile nerve block by xylocaine 1 month ago. The procedure had been completed without any intraoperative complications, but 2 days after circumcision, priapism developed and did not respond to decompression attempts with pharmacologic treatment, and glandulocavernous shunt was needed in the first 12 hours after development of the priapism.

Results

The ischemic event developed almost 3 weeks after circumcision and priapism. In the family history revealed no bleeding disorder, positive history of tissue necrosis of any body part, or recurrent deep vein thrombosis. On the physical examination, the skin over the shaft of the penis, the mucosal surface over the glans penis, and the surrounding skin over the pubic area were necrotic and tightly encircling the shaft like an eschar tissue.

All values of routine blood count and blood biochemical analysis were within normal limits. Further hematologic studies, including prothrombin time, International Normalized Ratio (INR), partial thrombin time, fibrinogen, homocysteine, and lipoprotein levels, revealed normal values. Antiphospholipid antibodies (both Immunoglobulin M (IgM) and Immunoglobulin G (IgG) types) were found to be negative. Factor VIII, antithrombin III levels, protein C level, and protein C activity were measured to be within normal limits. Free protein S level, however, was measured as 62.2 (70–130), which was lower than normal limits. Free protein levels of his mother and father were found to be within normal limits.

Only minimal debridement of the eschar tissue, surrounding the penis shaft and glans penis, was performed to restore the circulation to the viable cavernous bodies. Additionally, therapy with intravenous heparin infusion, antiaggregant therapy with low-dose salicylic acid and high molecular weight dextran had been initiated to augment the microcirculation. Two more operations for the debridement were performed before grafting the viable cavernous bodies of the penis. In each operation, special care was taken to preserve any viable tissue to end up with as much phallus as possible. Following the preservation of almost 2/3 of the cavernous tissue viable with conservative management, suprapubic area was closed with primary intention (Figure 2). Penis shaft was covered with split-thickness skin graft. The tip of the phallus was covered with buccal mucosa graft to make it like a glans tissue with different color (Figure 3). The patient has 4 cm long penis looking like almost normal with minimal tilt anteriorly due to ventral scar tissue. Family reported nocturnal erections are present.

Discussion

Hypercoagulable states, as a general term, refer to a broad spectrum of constitutional and acquired conditions that are known to be associated with

Figure 2 Appearance of the genital area of the patient after debridement of the eschar tissue. Defective surface on the suprapubic area and on the scrotum was closed by primary intention and healed without any complication.