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3 CIRCUMCISSION EXANGUINATION 4 ABSTRACT 5 A one month old male infant was circumcised by a non trained but long standing local 6 circumcision practitioner. He bled profusely and died after 8 hours post circumcision. 7 Autopsy findings were those of billary atresia. 8 Keywords :- circumcision, Billary atresia 9 CASE PRESENTATION 10 A 4 weeks old male delivered by a traditional birth attendant, was circumcised by an 11 experienced auxiliary nurse (the only person performing such in a local village).

Four hours 12 later, the child represented with complaints of continued bleeding from the site of 13 circumcision. She applied vitamin K injection and penicillin ointment topically and sent the 14 baby home. Approximately 3-4 hours later the baby was brought back to her dead. 15 Important history gotten at the autopsy venue (first contact of the parents with a medical 16 doctor since the child's delivery) were as follows: the child was delivered at 9 months 17 (probably at term), the circumcision was delayed for 3 weeks because around 7 days of age, 18 "the child was not strong enough" (in the words of the mother), was the 4th child in a family 19 of 2 boys and 2 girls, the elder brother did not bleed much after circumcision and the stool 20 was said to be pale coloured (non pigmented) in recent times. At autopsy, an apparently 21 healthy looking male infant was seen.

The head circumference was 34cm, mid arm 22 circumference was 14cm and the length was 1.9ft. The baby was moderately jaundiced and UNDER PEER REVIEW 23 the glands penis and site of circumcision was clean (no haematoma). All the organs were 24 present, well formed and in their normal anatomical positions.

The organs were very pale and 25 there was no excessive fluid collection in any of the serosal spaces. The lungs were 26 hyperventilated (with prominent rib markings), while

the liver (weighed 140g) had a smooth 27 shiny greenish surface with a homogenous greenish cut surface (figure 1a-c). 28 Microscopically, brain, heart, kidney, intestines and lungs showed features of 29 hypoxic/ischaemic injury.

Sections of the liver showed bile duct proliferation, mixed 30 inflammatory cell infiltration of the portal tract, grade 2 bridging fibrosis and bile plugging 31 (features of biliary atresia (BA). The autopsy diagnosis was concluded as massive 32 haemorrhage (hypovolaemic shock) post circumcision due to BA and defective hepatocyte 33 function.

34 LITERATURE REVIEW 35 Male Circumcision is one of the oldest and one of the most commonly performed surgical 36 procedures in practice today. It is commonly practiced and will continue to occur for 37 religious, cultural and medical reasons.¹⁻³ Complications of circumcision are broadly 38 classified into early and late complications.

Early complications include; bleeding (most 39 common), pain, inadequate skin removal, and surgical site infection. Others include chordee, 40 iatrogenic hypospadias, glanular necrosis, and glanular amputation. Post circumcision 41 bleeding in patients with coagulation disorders can be significant and sometimes even fatal.

2,4 42 Late complications of circumcision include; epidermal inclusion cysts, suture sinus tracts, 43 chordee, inadequate skin removal resulting in redundant foreskin, penile adhesions, phimosis, 44 buried penis, urethrocutaneous fistulae, meatitis, and meatal stenosis.²⁻⁴ Bleeding remains the 45 commonest complication encountered during and after circumcision.⁴

In most cases of 46 bleeding, which is usually minor, all that is required to achieve haemostasis is gentle pressure 47 on the area.² Excessive bleeding may be due to anomalous blood vessels or presence of UNDER PEER REVIEW 48 bleeding disorders.⁵ In situations where gentle pressure fails, circumferential bandage may be 49 used to aid haemostasis.²

Pharmacological agents like 1:100, 000 adrenaline can also be used. 50 Diathermy or suturing may be used for obvious bleeding vessels.² There are very few 51 documented cases of exsanguination following circumcision. Death following circumcision is 52 a very rare occurrence, such that in a series of 500,000 consecutive circumcisions in New 53 York City, no single fatality was recorded.⁶ 54 Biliary atresia (BA) is the single most frequent cause of death from liver disease in early 55 childhood.⁷

BA is the complete or partial obstruction of the lumen of the extrahepatic biliary 56 tree

within the first 3 months of life, that is characterized by progressive inflammation and 57 fibrosis of intrahepatic or extrahepatic bile ducts.⁸ BA mainly involves extra hepatic ducts, ⁵⁸ though intra hepatic ducts can also be involved and is usually seen in about 1 in 10,000 ⁵⁹ births.⁹

There are two major forms of biliary atresia (fetal and perinatal), based on the 60 presumed timing of luminal obliteration.⁷ ⁶¹ The etiology of BA is not clear, though toxic, infectious and autoimmune factors have been ⁶² implicated.¹ Clinically, BA is mainly seen in females. The infants are usually delivered at full ⁶³ term and jaundice develops during 2nd to 3rd week, while the stool colour is usually acholic.

¹ ⁶⁴ The index case though a male, was delivered at term, was jaundiced (onset not known) and ⁶⁵ passed pale coloured stool (acholic). The fetal form is a rare form and usually associated with ⁶⁶ other anomalies like interrupted inferior vena cava, polysplenia, congenital heart disease and ⁶⁷ malrotation of abdominal visera. It is seen in about 20% of cases. The suspected cause is ⁶⁸ aberrant intrauterine development of the extrahepatic biliary tree.

The perinatal form of BA is ⁶⁹ commoner, in which a presumed normally developed biliary tree is destroyed following ⁷⁰ birth.¹⁰ The main cause of BA is unknown, though autoimmunity, viral infections (reovirus, ⁷¹ rotavirus, and cytomegalovirus) and toxic factors are known to play roles.^{9,10} The index case UNDER PEER REVIEW ⁷² is probably a perinatal form BA, because the features listed above for fetal form were not ⁷³ seen.

⁷⁴ Grossly, the liver surface is smooth and homogenously green. The case in study was same as ⁷⁵ shown in figure 1a. Micro morphologic features of BA which are evident in about two thirds ⁷⁶ of cases are marked bile ductular proliferation, portal tract edema and fibrosis, inflammation ⁷⁷ of the hepatic or common bile ducts, parenchymal cholestasis and ⁷⁸ progressive destruction of the intrahepatic biliary tree.

If BA is unrecognized or uncorrected, cirrhosis develops within 3 ⁷⁹ to 6 months of birth.^{7,11} These features were characteristically seen in the index case with ⁸⁰ some bridging fibrosis. (Figures 2a &b, 3) ⁸¹ Clinically these infants appear almost normal at birth with normal birth weight and postnatal ⁸² weight gain.

Initially they pass normal coloured stools in the early days of to life before it ⁸³ progresses to acholic stools as the disease evolves. There is a slight female preponderance.^{7,9} ⁸⁴ The index case though a male, was claimed to be normal at birth by parents, though birth ⁸⁵ weight was not measured and passed normal coloured stool (yellowish brown) initially . ⁸⁶ Investigations that aid diagnosis include: elevated

hyperbilirubinemia, moderately elevated 87 aminotransferase and alkaline phosphatase levels.^{7,9} A combination of liver biopsy and ⁸⁸ radiologic excretion scintigraphy are the best investigations to confirm a diagnosis of BA.

BA ⁸⁹ is excluded if small bowel is visualized after using isotope ^{99m}Tc-DISIDA (di- ⁹⁰ isopropyliminodi-acetic acid). Ultra sound scan technique is also a good guide. In BA it ⁹¹ usually shows hypoplastic gallbladder, a cystic cavity at the porta and feature of established ⁹² cirrhosis early in life.¹² No investigation was done in the index case.

⁹³ Treatment of choice is hepatoportoenterostomy (kasai procedure) ie connecting the bowel ⁹⁴ lumen with the porta hepatitis, or else patients die from liver cirrhosis before 2 years of age.⁷ ⁹⁵ The success of this procedure is dependent on the level and variability in the anatomy of BA.⁷ UNDER PEER REVIEW ⁹⁶ For those that may not benefit from Kasai procedure, the other alternative is liver ⁹⁷ transplant.^{7,9} ⁹⁸ It was not seen in literature were circumcision lead to death in a BA patient, because in such ⁹⁹ circumstances, the patients must have been stabilized and proper precautions taken. The ¹⁰⁰ index case had no antenatal care, was not delivered in a hospital and never presented to a ¹⁰¹ hospital or qualified health worker.

In conclusion the post circumcision exsanguination seen ¹⁰² in this case was probably due to reduced / impaired hepatic functions with compromised ¹⁰³ production of clotting factors. Traditional / untrained circumcision practitioners should be ¹⁰⁴ informed of symptoms and signs (especially easily appreciated ones) that when seen, should ¹⁰⁵ discourage them from circumcising a male child. ¹⁰⁶ ¹⁰⁷ ¹⁰⁸ Figure 1: A) The homogenously green coloured liver with a smooth and shiny surface.

B) ¹⁰⁹ The inferior surface showing hypoplastic gallbladder. C) Hyper inflated lungs more marked ¹¹⁰ on the left lung. UNDER PEER REVIEW ¹¹¹ ¹¹² Figure 2: A and B shows early bridging necrosis between portal tracts and parenchymal ¹¹³ cholestasis ¹¹⁴ ¹¹⁵ ¹¹⁶ Figure 3: Showing marked bile ductular proliferation, portal tract edema and fibrosis, and ¹¹⁷ parenchymal cholestasis UNDER PEER REVIEW

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