CASE REPORT

Comparison of Antenatal and Postnatal Findings of Body Stalk Anomaly

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Abstract

Background: Body stalk anomaly was a rare, sporadic defect in the abdominal wall with the expulsion of the contents of the thoracoabdominal organs. Body stalk anomaly was reported in about one in 7,500 births within 10–14 weeks of gestation. Body stalk anomaly was a rare malformation with a prevalence of about 0.12 cases per 10,000 births (both live and stillbirth).

Aim: To compare the antenatal and postnatal findings of body stalk anomaly.

Case description: A 29-year-old multigravid patient was referred to our hospital due to a congenital anomaly. There were several findings on ultrasonography and magnetic resonance imaging (MRI), namely low-set ears, banana sign, neural tube defects, severe scoliosis, short umbilical cord, fetus attached to the placenta, abdominoschisis, thoracoschisis with some organs out, and defects in both legs. Hysterotomy was performed at 24 weeks of gestation. A male baby weighing 650 g with a body length of 33 cm and head circumference of 24 cm was born. His APGAR score was 1/1/1 and survived for 30 minutes after birth. The postnatal examination of the baby, performed postmortem, confirmed the antenatal diagnosis.

Synopsis: A case about a comparison between ultrasound and MRI prenatal screening with postoperative findings in the diagnosis of body stalk anomaly.

Conclusion: Antenatal ultrasonography provided an accurate diagnosis of body stalk anomaly, and the results could be a consideration for detection of the defect earlier. Furthermore, it could help in patient counseling about poor outcomes in neonates and termination planning earlier to avoid other additional risks or even complications of delivery.

Keywords: Antenatal, Body stalk anomaly, Diagnosis, Magnetic resonance imaging, Ultrasound.

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Body stalk anomaly was a rare, sporadic defect in the abdominal wall with the expulsion of the contents of the thoracoabdominal organs.[1] Body stalk anomaly was reported in about one in 7,500 births within 10–14 weeks of gestation.[2] Body stalk anomaly was a rare malformation with a prevalence of about 0.12 cases per 10,000 births (both live and stillbirth).[1,3]

The exact etiology of body stalk anomaly was unknown; three possible causes have been hypothesized, namely premature rupture of membranes, vascular compromise, and embryonic dysgenesis.[1]

Van Allen et al. proposed that a diagnosis of BSA must meet at least two of the following three criteria, exencephaly/encephalocele with a facial cleft, thoracoschisis and abdominoschisis (midline defect), and limb defect (e.g., clubfoot, polydactyly, oligodactyly, syndactyly, brachydactyly, or amelia).[2,4]

A 29-year-old multigravid patient was referred to our hospital due to a congenital anomaly. From anamnesis, she denied any history of consuming teratogenic drugs, infection during pregnancy, diabetes mellitus, premature rupture of membrane, previous congenital anomaly, and family history of congenital anomaly. Based on physical examination, the fundal height was palpable at the umbilicus (20 cm) with a normal fetal heart rate. There were several findings on ultrasonography, namely low-set ears, banana sign, neural tube defects, severe scoliosis, short umbilical cord, fetus attached to the placenta, abdominoschisis, thoracoschisis with some organs out, and defects in both legs (Fig. 1).

MRI result was similar to US findings. It showed a single live fetus intrauterine, the fetal position was bent with the head on the left inferolateral, and there were two superior and inferior extremities formed. The head surface of the gyrus appeared wide and smooth (lissencephaly). The ventricles appeared dilated asymmetrically, and there was an inhomogeneous intensity lesion in the left thalamus region which was suspicious for an intracranial mass. The right and left auricles appeared low (low-set ears). There are 2 lungs formed, positioned in the thoracic cavity. There were defects in thoracoabdominal organ followed by heart, intestines, and outside the thoracoabdominal cavity. The placenta was seen on the right anterolateral side, and the umbilical cord appeared short in single entanglement (Fig. 2). Laboratory examination showed normal IgM and IgG toxoplasma, IgM rubella, IgM CMV, IgM HSV I, IgM HSV II,

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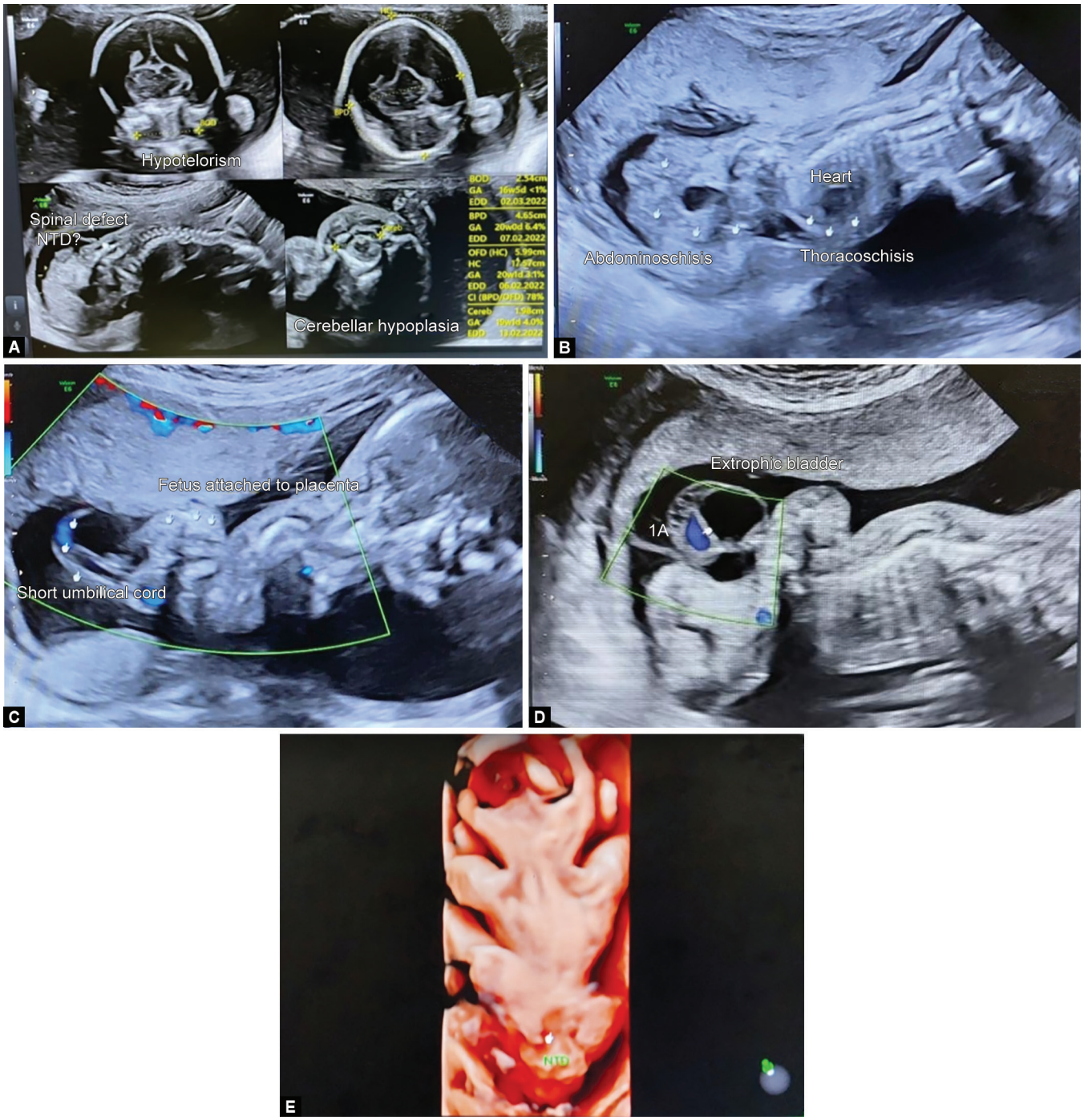
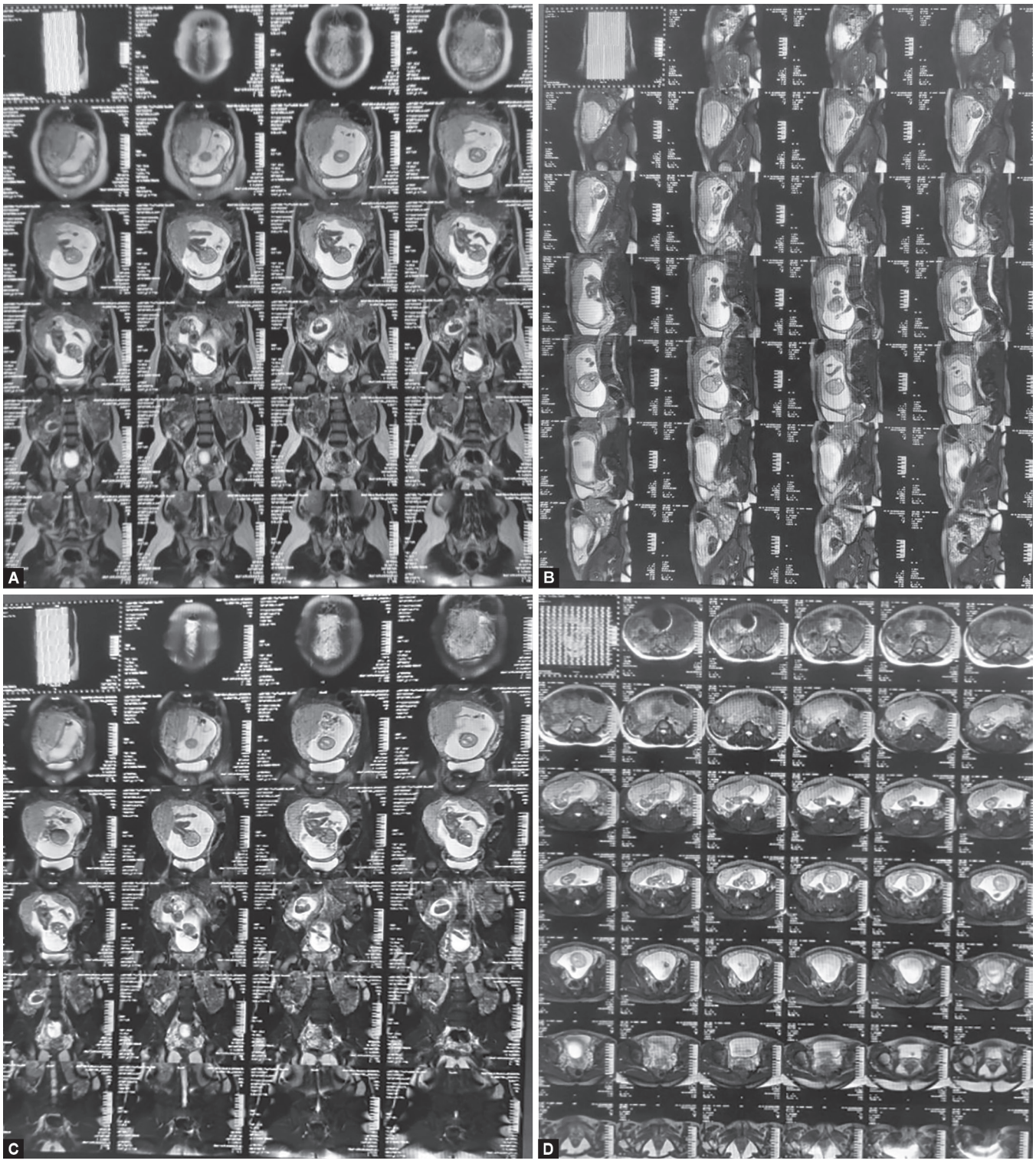
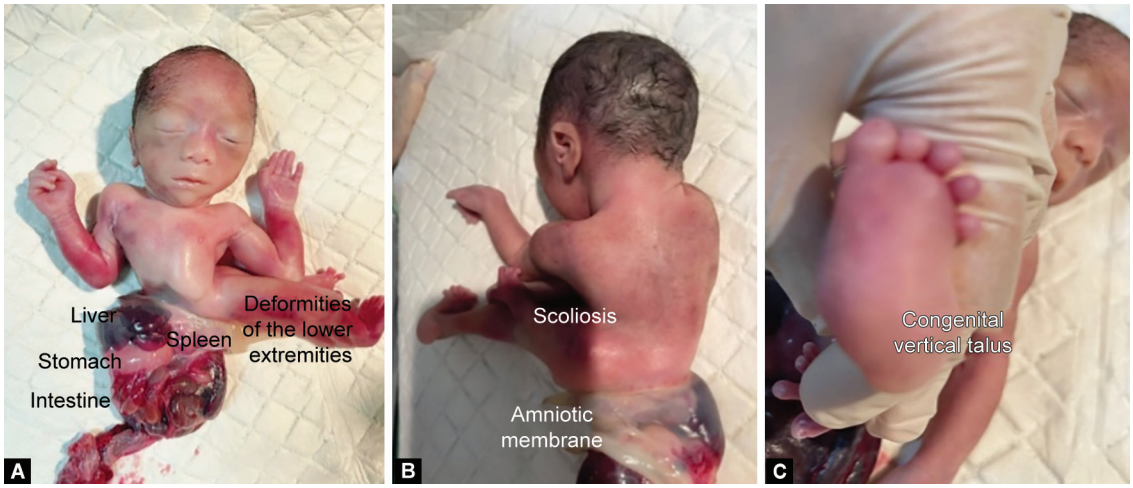


Figs 1A to E: Ultrasound examination showed (A) Hypotelorism, neural tube defects, a banana sign indicating cerebellar hypoplasia; (B) Thoracoschisis and abdominoschisis; (C) Fetus attached to the placenta and short umbilical cord; (D) Extrophic bladder; and (E) 3D showing neural tube defects and scoliosis



Figs 2A to D: Magnetic resonance imaging examination showed a single live fetus intrauterine, the position of the fetus was bent with the head on the left inferolateral. There were 2 extremities superior and inferior formed. The head surface of the gyrus appeared wide and smooth (lissencephaly). The ventricles appeared asymmetrically dilated, there was an inhomogeneous intensity lesion in the left thalamus region, and an intracranial mass was suspected. Right and left auricles looked lower (low set ears). There were 2 lungs formed, positioned in the thoracic cavity. There were thoracoabdominal defects followed by the heart, intestines and outside the thoracoabdominal cavity. The placenta was seen on the right anterolateral side, the umbilical cord appeared short in 1 circular tie



Figs 3A to C: Postoperative male neonate weighing 650 g was born. There were abdominoschisis, lower limb deformity, scoliosis, and congenital vertical talus

IgG HSV II. However, there was increased IgG Rubella, IgG CMV, IgG HSV I, but they were not significant as an acute infection markers.

We conducted a joint conference with the ethical committee, religious leader, legal adviser, and clinicians from multidiscipline. From the joint conference, we agreed to perform hysterotomy due to the poor fetal prognosis (lethal anomaly). After we did informed consent and choice to the patient and her family, we did hysterotomy at 24 weeks of gestation.

A male baby weighing 650 g with a body length of 33 cm and head circumference of 24 cm was born. His APGAR score was 1/1/1 and survived for 30 minutes after birth. The postnatal examination of the baby performed postmortem, confirmed the antenatal diagnosis (Fig. 3). There were abdominoschisis with scoliosis and congenital vertical talus. Postmortem pathological analysis could not be performed. In addition, the results of postoperative laboratory examinations showed an increased alpha-fetoprotein 918.56 ng/mL. Although the recurrence rate of body stalk anomaly remained low, we still suggested her for having multivitamin, such as high dose folic acid, vitamin D, and other antioxidants every day at least for 6 months before the next conception for preventing the recurrence of body stalk anomaly in the subsequent pregnancy. Furthermore, she should have routine antenatal care and do some screening in every trimester of her pregnancy with obstetricians.

Antenatal ultrasonography and MRI provided an accurate diagnosis of body stalk anomaly and the results could be a consideration for detection of the defect earlier. However, it depended on the operator's skill, knowledge, and experience. Furthermore, it could help on patient counseling about poor outcomes on neonates and termination planning earlier to avoid other additional risks or even complications of delivery.

Author Contributions

Peby Maulina Lestari and Yurizka Sabrina contributed to the acquisition of data. Wim T Pangemanan, Nuswil Bernolian, and Putri Mirani designed and drafted the article. Ahmad Kurdi Syamsuri and Abarham Martadiansyah were responsible for critical manuscript revision. M Hatta Ansyori and Cindy Kesty were responsible for the final approval of the version to be submitted.

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