Case report: sacral parasitic twins

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Summary

Introduction: Sacral parasitic twins originate from one fertilized ovum and they have one placenta and the same sex. *Case Report:* A 23-year-old woman was referred to our clinic. Examination by touch revealed a mass that was in the sacral region but the borders could not be fully examined. The solid mass, which was conjoined to the sacrum, had a soft texture. The infant's appearance was macroscopically normal. When the mass was examined by palpation, there were structures which felt like extremities. The mass was 20 x 11 x 9 cm in size. *Conclusion:* The differential diagnosis should include sacrococcygeal teratoma. In our case the differential diagnosis was done by histopathologic findings. This case, which involved a tumoral formation at the sacral region in the antenatal period, was detected during delivery. A sacral parasite is a rarely seen phenomenon and as such the diagnostic information of this case could be useful.

Key words: Parasitic twin; Teratoma; Ultrasonography.

Introduction

Sacral parasitic twins originate from one fertilized ovum and they have one placenta and the same sex. The incidence of this condition has been documented as being 1/40,000-1/200,000 live births [1]. Females are affected more frequently than males by a ratio of three to one. Compared to the USA it is seen in India and Africa more frequently [2]. After the 13th day following fertilization, twins should start to split. It is assumed that the most important factor that underlies formation of conjoined twins is the failing of this complete separation. Conjoined twins are classified according to their conjoined body parts [1, 2].

Parasitic twins (heteropagus) are asymmetric conjoined twins. The parasitic twin is completely dependent on its twin and is smaller and less likely to the organism. It is difficult to deliver such patients. A review of the literature reveals 28 cases reported thus far. We present this rare occurence.

Case Report

A 23-year-old woman was diagnosed as having a "twin pregnancy with pain" in another center and was referred to our clinic. Her gravidity was three, parity was two and number of live children was two. Antenatal care or ultrasonography was not performed. The patient was referred to our clinic urgently because labor had started. There was one fetal cardiac activity. At the patient's vaginal examination the cervix was completely effaced and dilatated. The head descent by station was +2, and heart beat sounds were positive. After the head, the anterior and posterior shoulders were delivered. At the umbilicus level the fetus could not be delivered so controlled fraction was applied. The McRoberts and Rubin's maneuvers were attempted but failed. Examination by touch revealed a mass that was in the sacral region but the borders could not be fully examined. The heart rate of the fetus was descelerated. Surgery was initiated immediately. The Pfannenstiel incision was performed to access the abdomen and a lower uterine incision was made. A 4,200 g, 50 cm in length female fetus was delivered by cesarean section and the Apgar score of the infant was 0. Postoperative resuscitation was carried out with no results. Atony, bleeding or other maternal complications were not seen.

The solid mass which was conjoined at the sacrum had a soft texture (Figure 1). The infant's appearance was macroscopically normal. When the mass was examined by palpation, there were structures which felt like extremities. The autopsy revealed that the fetuses anus opening was displaced to the left because of the sacral mass. A mass 20 x 11 x 9 cm in size which had continuity with the fetal skin in the sacrococcygeal region was documented. The outer surface of the mass was smooth and had a partly lobulated contour. At the surface of the mass, organoid-like tissues as bone, cartilage and fatty tissue (extremity parts) were detected (Figure 2).

Discussion

Prenatal ultrasonography (US), echocardiography, and 3-dimensional magnetic resonance imaging (MRI) which generally gives detailed information about conjoined twins, also helps in deciding whether the pregnancy should continue or be discontinued [1, 2]. Gestations that cannot be separated and have common important anomalies have to be ended. Conjoined twins can be recognized even at the 23rd gestational week by US. The presented case was not properly followed and antenatal diagnosis was not performed, thus appropriate treatment was not carried out [3, 4].

Systematic classification of conjoined twins was first established by Schwalbe *et al*. in 1905 [5]. The differential diagnosis should include sacrococcygeal teratoma. Teratoma is usually accompanied by malignancy. In our case the differential diagnosis was done by histopathologic findings. For this reason teratoma is considered as

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Fig. 1

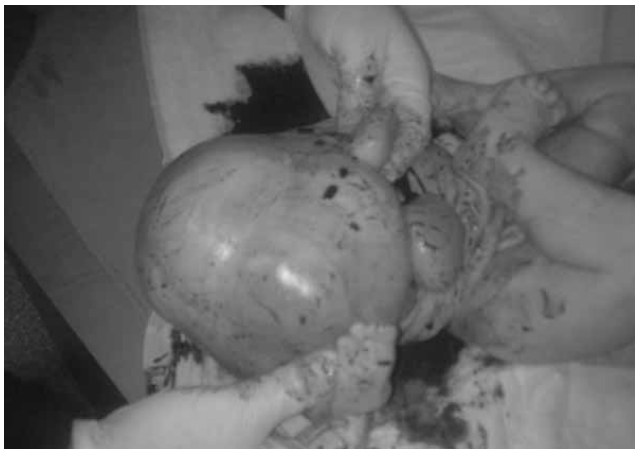

Fig. 2

Figure 1. — A 20 x 11 x 9 cm mass which has continuity with fetal skin in the sacrococcygeal region.
Figure 2. — At the surface of the mass organoid-like tissues as bone, cartilage and fatty tissue (extremity parts) were detected.

a different concept from a parasitic twin. Teratoma consists of internal organs which are at different developing stages [6-9]. In conjoined twins DNA typing and karyotype analysis studies have been done. Spencer *et al*. reported that parasitic twins were always monozygotic [8]. We do not have the facilities for these analyses, thus they are not performed. For a female who has a parasitic mass and the surface tissue of the mass looks like scrotal tissue, benefits of DNA typing could not be underestimated.

Spencer *et al*. reported 20 parasitic cases, 18 of which were localized and two with extensive conjunctions. All of these cases were medial to the dorsal midline [6, 8]. A literature review revealed eight more cases [3, 7, 9, 10]. When all reported cases were reviewed it was found that 18 of the cases were females and five were males, including our case; six of the case sexes could not be detected [6]. Anatomy and morphology of the reported parasites were related to the affected site. Upper extremities and related bones are seen in parasitic twins which are localized at the cervical and upper thoracic region. Lower extremities and bones are seen in parasitic twins which are localized at the lumbar and dorsal region [10]. In a parasitic mass, existence of polymorphic tissue belonging to a multi-organ system supports the theory established by Spencer; this theory suggests that these lesions are aborted or parasitic twins [8].

There are studies reporting that benign teratoma and lipomatosis tissues were detected in a parasitic mass [10]. This condition shows that there is a thin line between duplication aborted by teratoma, fetal inclusion, many types of parasitic twins and conjoined twins. When all reported cases were reviewed it was noted that cesarean section was performed in all cases because of dystocia.We attempted a vaginal delivery at first but had to resort to cesarean section.

This case involved a tumoral formation at the sacral region in the antenatal period which was detected during delivery. After histopathologic examination it was revealed that the mass was a sacral parasite (pigopagus parasiticus) and definitely different from teratoma. This case is presented to share diagnostic information as sacral parasite is a rarely seen phenomenon.

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