Gluteus as a rare localization of extragonadal teratoma

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To the Editor,

Teratomas represent the most common germ cell tumors in children[1]. They can be gonadal, more common in adolescents, or extragonadal, primarily in neonates and young children. Teratomas develop from totipotent primordial cells and may originate anywhere along the midline. Common sites for extragonadal teratomas are the sacrococcygeal region, which accounts for 35-60% of all teratomas, the mediastinum, the retroperitoneum, the head and neck and the central nervous system[2–4]. Other localizations are rare, especially non-midline ones that are very often lateralized expansions of midline teratomas, such as those arising from sacrococcygeal region.

We here describe the case of a newborn girl presenting with a gluteal mass, that revealed to be a primary extragonadal teratoma. At birth she presented with a hard-elastic, mobile and painless mass localized within the right gluteus (Fig.1), that had not been noted on prenatal ultrasound. At two days of life, an echography was performed, revealing a subcutaneous irregularly hypoechoic mass with fluid areas inside and small vessels, with aspecific characteristics. The dimension of the mass was 24x15 mm and rapidly increased in size, reaching 40x25mm, and in the number of fluid areas (Fig. 1). Alfafetoprotein serum concentration resulted 4586 ng/mL (refence value at the 2 week-1 month interval at which she was tested 316-6310 ng/mL) and hCG was 0,8 UI/L (normal value <5 UI/L)[5]. After performing an MRI that excluded other lesions, the mass was removed and a biopsy was performed, revealing an immature teratoma, grade 3 according to

Norris's classification. Since extragonadal teratomas out of the midline are very rare, she has been followed up thoroughly for 3 years with regular periodic blood tests and radiological assessments, but no other primary lesions or recurrencies were found.

Teratomas can be malignant (12-14%) or benign, further divided into mature (50-60%) and immature (18-34%). Immature teratomas contain fetal tissue, most often neuroectodermal, the amount of which is scored according to a grading system introduced by Norris. Grade 3 is that with most neuroectodermal tissue, and have an increased incidence of local recurrence and malignant degeneration[3,4,6]. Complete and prompt surgical resection is the gold standard for definitive therapy in benign teratomas, both mature and immature[4,6].

Teratomas develop along the midline because they originate from the incomplete differentiation of totipotent primordial cells that arise in the yolk sac and migrate along the mesentery to the gonadal ridge during the 4^{th}-5^{th} week of embryologic development[3,4]. Indeed, most of the gluteal teratomas reported in literature are lateralized sacrococcygeal teratomas with a connection to the coccyx, since sacrococcygeal teratomas are thought to be derived from totipotent cells of the Hensen's node (primitive knot), an area at the cranial end of the primitive streak[7,6].

Other authors reported rare sites for lateralized teratoma development such as kidney, liver and temporozygomatic region[8,9]. Rare lateralized extragonadal localizations should not mislead the clinical suspicion of teratomas, and a primary localization should always be excluded. Taking also the patient's age into account, it can sometimes be considered to perform a PET scan. To the best of our knowledge, there is just one case in literature of a gluteal teratoma not in connection with the coccyx, as in our patient, thus confirming the possibility of this very rare localization[10]. The biological mechanism for germ-cell migration in such anatomical regions is still to be elucidated.

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CONFLICT OF INTEREST

The authors declare that there is no conflict of interest.

ETHICS STATEMENT

Written informed consent has been obtained from the patient to publish this paper.

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LEGENDS

Figure 1: A Picture of the gluteal mass, B MRI image of the immature teratoma