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Case Report

Haematopoietic bone marrow and bony trabeculae: An uncommon mesodermal derivative in mature teratoma in a 14 year old girl

Priya Kumari¹, Rudra Panigrahi², Premanand Panda³, Pragnya Paramita Mishra^{1,*}

¹Dept. of Pathology, Hitech Medical College, Rourkela, Odisha, India

²ESIC Model Hospital, Rourkela, Odisha, India

³Dept. of Radiology, JP Hospital & Research Centre, Rourkela, Odisha, India



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ABSTRACT

Teratomas are embryonal tumours that emerge when totipotent germ cells evade the primary organisers' control over their growth and give rise to tumours that contain tissue from all three germ layers. They are cystic or solid, with mature or immature components. Histologically, teratomas are classified as mature (benign in 95% of cases) and immature with malignant transformation. They are rare tumours with a frequency of 1 in 10,000 births. Teratomas are located more often in the sacrococcygeal region, and in the ovary, the cervical localization represents 1.5% to 5% of all the localizations. They predominate among females. Mesodermal derivatives are relatively rare, and solitary organic development, especially of a mesodermal derivation in a random teratoma, is unique and crucial to distinguish for clinical reasons. Here, we present a 14-year-old female who presented with a submental swelling that, on histopathology, was diagnosed as a mature teratoma with hemopoietic tissue and bony trabeculae.

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1. Introduction

“Teratoma” is historically derived from the Greek word “Teraton, which means a monster, and the word was coined by Virchow in 1863.¹ Teratomas are tumours that frequently contain a variety of cell types from each of the three germ layers. Teratomas have been reported to occasionally contain more complex organs in addition to hair, teeth, and bone. Although the precise cause of teratomas is unknown, the most frequently accepted explanation postulates that they may have originated from the first germ cells. Teratomas can be solid and malignant (immature), or they might be benign, well-differentiated cystic lesions. Head and neck teratomas are rare benign tumours, accounting for 3% of all teratomas. Mesodermal derivatives in teratomas are relatively uncommon. Again,

hemopoietic tissue is also an unusual mesodermal element in teratomas, and only 0–9.5% of mature teratomas develop this component.^{2,3} On the other hand, the presence of well-developed hemopoietic bone marrow with intercepting osseous trabeculae represents organic development, which has only been described in ‘fetiform teratoma’ or ‘homunculus’.⁴ Herein, we discuss a case of mature benign teratoma with uncommon mesodermal derivatives in the form of hemopoietic bone marrow with intercepting osseous trabeculae in a 14-year-old female in the head and neck region.

2. Case Report

A 14-year-old female presented to ENT OPD of Model Hospital with a long-standing history of midline anterior neck swelling below the chin since birth, but the swelling has increased in size since the last 4 years. The swelling

* Corresponding author.

E-mail address: pparamita1982@gmail.com (P. P. Mishra).

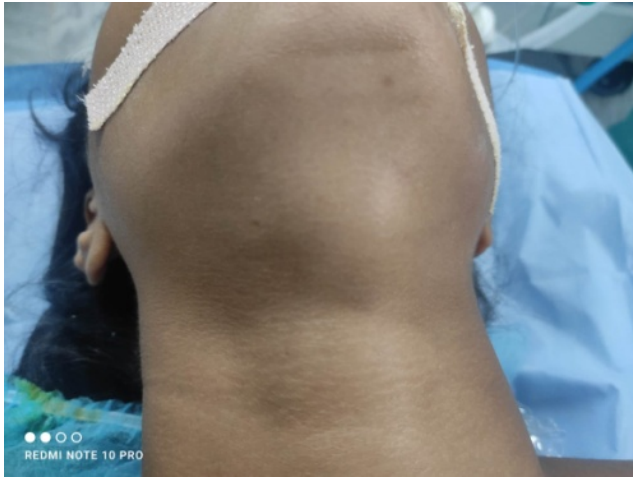


Fig. 1: Photomicrograph: Neck swelling

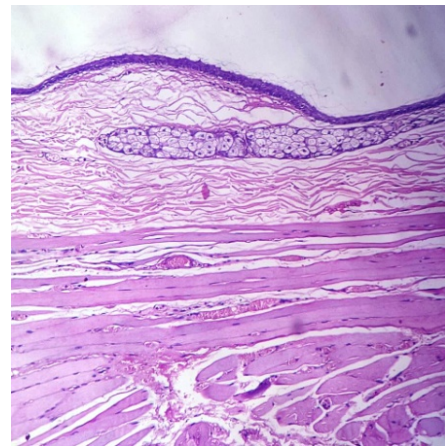


Fig. 4: Photomicrograph : H & E picture showing stratified squamous epithelium showing skin adnexal structure (10x)

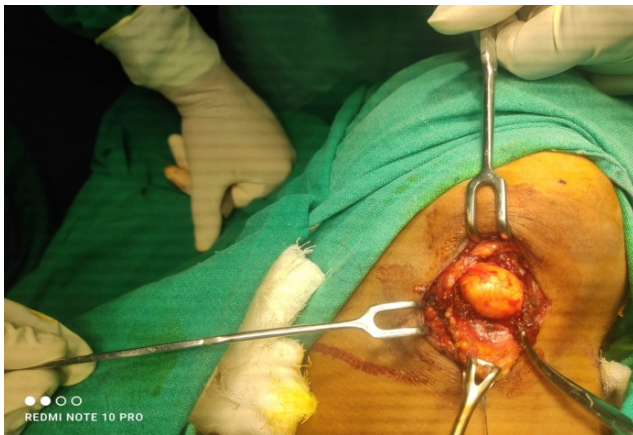


Fig. 2: Photomicrograph: Intra-operative finding

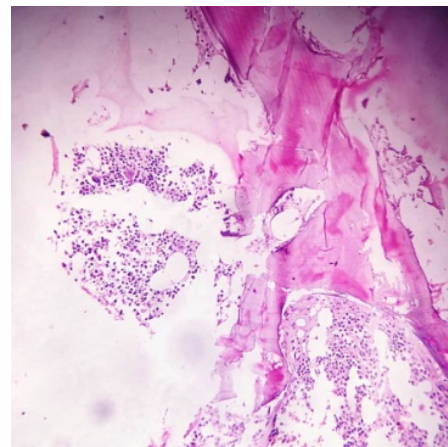


Fig. 5: Photomicrograph: H & E picture showing mature bony trabeculae with haematopoietic island (10x)

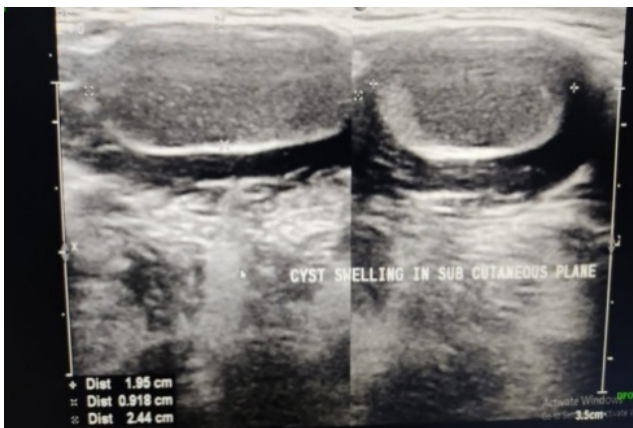


Fig. 3: Photomicrograph: Ultrasonography reveals cystic swelling with internal echoes in subcutaneous plane

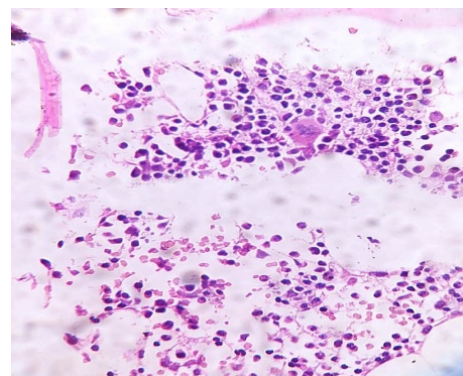


Fig. 6: Photomicrograph: H & E picture showing haematopoietic tissue along with megakaryocyte (40X)

was painless and measured 3.0 x 3.0 cm in size. She did not complain of any dysphagia, respiratory distress, or hoarseness. There was also no history of constitutional symptoms or hypo- or hyperthyroidism. In family history, the mother had hypothyroidism and was on medication. On examination, the neck swelling was soft in consistency, non-tender, and had a smooth surface without discoloration of the skin. The mobility of swallowing was not elicited but on tongue protrusion movement of swelling was elicited. The surrounding skin was healthy except for a small area showing minimal signs of inflammation. Fine needle aspiration cytology (FNAC) was performed, and the aspirate showed clusters of anucleated squames. With this, an impression of an epidermal inclusion cyst was made. To rule out other possibilities, Neck ultrasonography (USG) was performed, which showed a well-defined unilocular cystic lesion in the subcutaneous plane of the submental region with internal echoes.

A possible differential diagnosis of thyroglossal duct cyst was offered. With this diagnosis, a Excision of Midline Cyst was performed under GA, and the specimen was sent for histopathology. Grossly, a well-encapsulated globular tumour was received with cystic consistency, measuring 2.5 x 1.5 x 1.5 cm. The outer surface was unremarkable, and the cut surface shows whitish, cheesy material along with bony hard tissue. In microscopy, a cystic structure lined by squamous epithelium along with skin adenexal structure and a lumen containing keratin, cyst macrophages with mature bony trabeculae, and hemopoietic tissues were seen. Hematopoietic tissues include erythroblasts, myeloid series cells, and megakaryocytes. No thyroid tissue was identified in the sections studied. No malignant cells were seen. Features were suggestive of a mature cystic teratoma with bony tissue and hemopoietic elements. The patient recovered well post-operatively with no further complications to date.

3. Discussion

Teratoma is the most common germ cell tumour of the ovary. Other sites include the testis, central nervous system, and mediastinum. Children and women in their reproductive years are usually affected, but 25% of teratomas arise in postmenopausal women. Patients normally present with abdominal pain, heaviness, or exceptionally with abdominopelvic swelling and abnormal uterine bleeding. However, 25–60% of individuals experience an asymptomatic outcome. Preoperative diagnosis of mature teratomas can satisfactorily be accomplished on radiology, depending on the heteroechoogenicity exerted by its variegated tissue constituents, especially bone, cartilage, and teeth. In our case, the patient was 14 years old and complained about midline anterior neck swelling below the chin since birth, but the swelling has increased in size since the last 4 years.

Neck ultrasonography (USG) was performed and showed a well-defined unilocular cystic lesion in the subcutaneous plane of the submental region with internal echoes.

Teratomas mesodermal composition is largely responsible for their overall complexity. Adipose tissue, cartilage and bone are the common mesenchymal tissue found in about 66.7 - 78.6%, 21.4 - 38.6% and 11.9 - 18.6% of teratomas, respectively. Lymphatics and myxoid tissue are the least reported mesodermal tissues. Hematopoietic tissue is also a relatively secluded mesodermal constituent. It predominantly occurs as an isolated aggregate of hemopoietic precursors or as lymphoid tissue underlying pharyngeal, respiratory, or gastrointestinal mucosa. On extremely rare occasions, a well-formed spleen, lymph node, or trabecular bone marrow develops. However, despite thorough research, it was impossible to find in previously published literature on teratomas the unique presence of the hemopoietic bone marrow identified in the discussed article. The hemopoietic bone marrow is a separate organ, but its mere presence within an otherwise mature teratoma is quite uncommon.

In the future, new insights into embryogenesis or even parthenogenesis may be revealed through a thorough examination of teratomas for such organogenesis and their meticulous histological characterization.

4. Conclusion

We reported a rare case of teratoma in a 14-year-old girl in the head and neck region. Although uncommon, teratoma should be considered in the differential diagnosis of patients presenting with swelling in the head and neck. Careful clinical examination, radiological investigation, and histopathologic examination can greatly aid in pinpointing the diagnosis.

5. Conflicts of interest

There are no conflicts of interest.

6. Source of Funding

None.

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Author biography

Priya Kumari, Post Graduate

Rudra Panigrahi, ENT Specialist

Premanand Panda, HOD

Pragnya Paramita Mishra, Assistant Professor

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