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Extraskelatal Ewing sarcoma of the stomach: A rare case report

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Abstract

BACKGROUND

Extraskelatal Ewing sarcoma (EES) is a rare and highly malignant small round cell tumor associated with a poor clinical outcome. Ewing sarcoma (ES) involving the stomach is an uncommon presentation and can be easily confused with other small round cell tumors. We herein present a rare case of ES involving the gastric area.

CASE SUMMARY

We report a case of gastric ES in a 19-year-old female patient who initially presented with a complaint of a tender epigastric mass for 5 d. Contrast-enhanced abdominal computed tomography revealed a soft-tissue-density mass with a diameter of 8.5 cm between the liver and stomach; the mass was connected to the gastric antrum. Then, the mass was surgically excised completely. Upon histopathological, immunophenotype and molecular analysis, the mass was identified to be a primary gastric ES.

CONCLUSION

EES is an aggressive tumor with poor prognosis. Therefore, early diagnosis and timely intervention are essential for a good prognosis. It is imperative for us to raise awareness about this rare tumor. Surgical resection is still the best treatment option.

Key Words: Stomach neoplasms; Extraskelatal; Ewing's sarcoma; CD99; FLI-1; Case report

Core Tip: Ewing sarcoma (ES) is a rare and highly malignant small round cell tumor. Extraskelatal ES is more common in the paravertebral region, extremities, and retroperitoneum and very rarely occurs in the stomach. Due to its rarity, there are few studies on its clinicopathological characteristics and diagnostic protocols. It is an aggressive tumor with poor prognosis, and therefore, early diagnosis and timely intervention are vital for a good prognosis. Herein, we report on a case of primary gastric ES which was timely resected surgically. This was followed by chemotherapy, which was comprised of alternating vincristine–doxorubicin, cyclophosphamide, and ifosfamide–etoposide every 3 wk. Ultimately, the patient was tumor-free and achieved an excellent prognosis.

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INTRODUCTION

Ewing sarcoma (ES) is a rare and highly malignant small round cell tumor[1]. It is characterized by different degrees of neuroectoderm differentiation and an EWSR1-ETS fusion protein[2]. ES often occurs in the pelvis, long axial bones, and the femur, whereas extraskelatal ES (EES) is more common in the paravertebral region, extremities, and the retroperitoneum[3]. It is very rare for EES to occur in the stomach. As reported in Hopp *et al*'s study, the incidence of EES accounted for 15%-20% of all ES cases [4]. Among the 70 EES cases reported by Huh, only 4 cases (5.7%) were of primary gastric ES[5]. Due to its rarity, there are few studies on its clinicopathological characteristics and diagnostic protocols. We herein report a case of primary gastric ES to raise awareness about this rare tumor for clinicians and radiologists.

CASE PRESENTATION

Chief complaints

A 19-year-old female patient presented with a complaint of a tender epigastric mass for 5 d.

History of present illness

Five days ago, the patient was admitted for a complaint of a tender epigastric mass. There was no prior history of gastroesophageal reflux symptoms or black stools, and she recently reported upper abdominal fullness after intake of little food.

History of past illness

The patient had no history of past illness.

Personal and family history

No personal or family history was available.

Physical examination

On clinical examination, slight tenderness and an immovable mass in the right upper abdomen were identified.

Laboratory examinations

Serum fetoprotein, carcinoembryonic antigen, squamous cell carcinoma antigen, neuron-specific enolase, carbohydrate antigen 125, carbohydrate antigen 153, and carbohydrate antigen 19-9 were all normal.

Imaging examinations

The abdominal contrast-enhanced ultrasound revealed a hypoechoic mass with a diameter of 8.5 cm between the liver and stomach. It had poorly defined boundaries and an irregular shape and mutual continuity with the gastric antrum. The pancreas was compressed by the mass, and a branch of the

celiac trunk was seen to extend into the center of the mass. The ultrasound contrast agent SonoVue (12 mL) was injected through the cubital vein, and the mass and liver parenchyma showed synchronous homogeneous enhancement in the arterial phase; conversely, the mass showed non-homogeneous enhancement in the portal vein phase and slightly non-homogeneous enhancement in the hepatic delay phase. Contrast-enhanced ultrasound indicated a gastric stromal tumor (Figure 1). Contrast-enhanced abdominal computed tomography (CT) revealed a soft-tissue-density mass measuring 8.5 cm in diameter between the liver and stomach. The mass had uneven internal density, mild continuous enhancement in the arterial phase, and blood supply from a small branch of the gastroduodenal artery. Furthermore, the mass was connected with the gastric antrum. There were no clearly enlarged lymph nodes in the abdominal cavity or retroperitoneum. Contrast-enhanced abdominal CT indicates a gastric antrum tumor (Figure 2). Subsequent esophagogastroduodenoscopy revealed that the gastric antrum mucosa was intact and smooth (Figure 3). Chest CT and bone scan emission CT showed no abnormal findings.

FINAL DIAGNOSIS

According to the above history and the related imaging examination, the final diagnosis was of an abdominal tumor potentially of a gastric origin.

TREATMENT

The patient underwent distal subtotal gastrectomy, gastroenterostomy, intestinal anastomosis, and regional lymphadenectomy. The resected specimen contained a firm, round tumor with extraluminal growth. It was located in the gastric antrum and measured 8.5 cm × 5.0 cm. Histological examination revealed that the tumor developed from the serous layer of the stomach and involved the muscularis externa of the stomach (Figure 4A). Tumor cells were characterized by uniform, compact, round-to-oval nuclei, a modest amount of pale eosinophilic cytoplasm, and pseudorosette formation. There were punctate areas of necrosis, but the degree of necrosis was less than seen in usual small-cell carcinoma; atypical mitoses were occasionally observed (Figure 4A). No metastatic tumors were found in the regional lymph nodes. On immunohistochemistry, the tumor cells were found to strongly express CD99 on the membrane and in the cytoplasm, vimentin in the cytoplasm, Fli-1 in the nuclei, and NSE and SYN in the cytoplasm (Figure 4B-F). Molecular analysis using fluorescence *in situ* hybridization revealed the EWS-FLI1 gene translocation t(11; 22) (q24; q12). Therefore, the tumor was eventually determined to be primary gastric ES. The patient recovered and was discharged from the hospital. Two months after the surgery, she underwent chemotherapy which was comprised of alternating vincristine–doxorubicin, cyclophosphamide, and ifosfamide–etoposide every 3 wk.

OUTCOME AND FOLLOW-UP

Ultimately, the patient was tumor-free and achieved an excellent prognosis. At the time of writing this paper, she was still alive and undergoing her tenth round of chemotherapy, and she showed no sign of a relapse 11 mo after the surgery (Figure 5).

DISCUSSION

ES is a highly malignant and small round cell tumor. It comprises classic ES of the bone, EES, and peripheral PNETs of the bone and soft tissue[1]. Primary gastric ES is exceptionally rare. Czekalla *et al*[6] first reported a case of gastric ES in 2004, and few cases have been reported in the literature so far. We summarize the reported cases of primary gastric ES in Table 1, describing their clinical presentation, immunohistochemistry findings, treatment and outcome[1,4,6-15]. The rare occurrence of EES in the stomach may be related to its origin from primitive tumor cells. Although the histogenesis of ES is unclear, it is well known that the most common chromosomal rearrangement in ES is a translocation between chromosomes 11 and 22 [t(11;22) (q24; q12)], which can result in new EWSR1-ETS fusion proteins as transcription factors that regulate the target genes, consequently leading to cell transformation and generation of tumors with ES morphology and gene expression characteristics[16]. By projecting t(11; 22) (q24; Q12) translocation, Sole *et al*[17] generated a large number of immortalized cells (EWIma cells) resistant to EWSR1-FLI1 expression from primary mesenchymal stem cells; these cells induced tumors and metastases in mice, suggesting that ES originates from human bone marrow primitive mesenchymal cells. Therefore, embryonic stem cell tumor cells of ES are possibly derived from

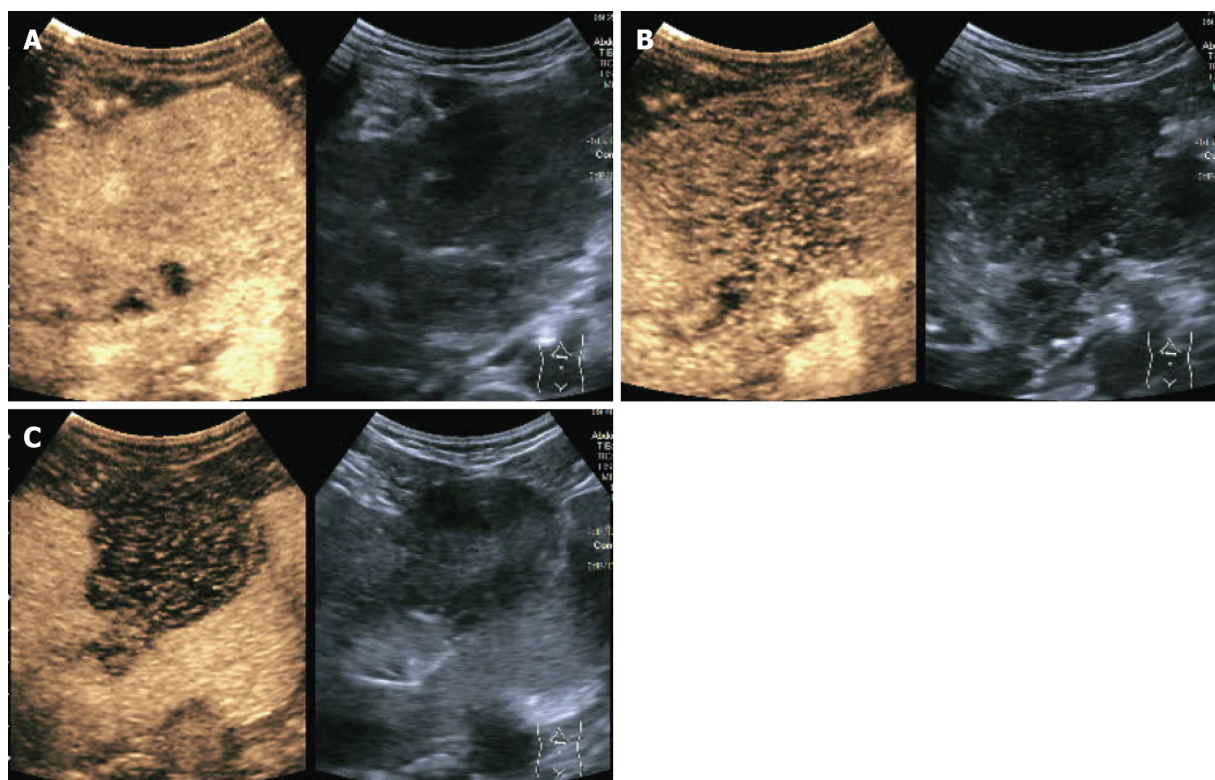
Table 1 Clinical characteristics and outcomes in reported cases of gastric Ewing's sarcoma

| Ref. | Age in yr, sex | Clinical presentation | Location | Tumor size in cm | Immunohistochemistry | Molecular genetic detection | Distant metastasis | Neoadjuvant chemotherapy | Surgery | Postoperative chemotherapy | Follow-up in mo | Outcome |
|-----------------------------------|----------------|-------------------------------------|---------------------|------------------|--|-----------------------------|------------------------|--------------------------|----------------|----------------------------|-----------------|---------|
| Czekalla <i>et al</i> [6], 2004 | 14, M | Anorexia, epigastric pain | Anterior wall body | 5 | CD99, CD117, S100, vimentin, neurofilament, NSE | t(11;22)(q24;q12) | Liver | Yes | SG | No | 24 | Alive |
| Soulard <i>et al</i> [7], 2005 | 66, F | Epigastric pain | Antrum | 8 | CD99, S100, NSE, vimentin | t(21;22)(q22;q12) | No | No | Gastrectomy | Yes | 10 | Dead |
| Colovic <i>et al</i> [8], 2009 | 44, F | Epigastric pain | Posterior wall body | 10 | CD99, S100, vimentin, NSE, PGP9.5 | NA | No | No | Excision | No | 20 | Alive |
| Rafailidis <i>et al</i> [9], 2009 | 68, M | Abdominal pain, dyspepsia, weakness | Body | 12 | vimentin, NSE, CD99, CD117, Leu-7 | t(11;22)(q24;q12) | Liver | No | SG | Yes | 13 | Dead |
| Inoue <i>et al</i> [10], 2011 | 41, F | Abdominal pain | Anterior wall body | 9 | CD99, vimentin, CD117, S100, chromogranin A, synaptophysin | t(11;22)(q24;q12) | Enterocoelia | No | DG | Yes | 110 | Dead |
| Kim <i>et al</i> [11], 2012 | 35, F | No special symptoms | Antrum | 5.5 | CD99, synaptophysin | NA | No | No | WR | No | 11 | Alive |
| Song <i>et al</i> [12], 2016 | 55, M | Upper abdominal pain | vomiting High body | 6.5 | CD99, FLI1, chromogranin | EWS-FLI1 fusion transcript | Lymph nodes | No | TG | Yes | 13 | Alive |
| Kumar <i>et al</i> [13], 2016 | 32, F | Epigastric pain | Lesser curvature | 11 | CD99, vimentin, S100 | NA | No | No | Excision | Yes | 12 | Alive |
| Khuri <i>et al</i> [14], 2016 | 31, F | Upper gastrointestinal hemorrhage | Lesser curvature | 11 | CD99, FLI1, vimentin, Ki67 | (11;22 translocation) | Pancreas Splenic hilum | No | TDPSLA | No | 36 | Alive |
| Maxwell <i>et al</i> [15], 2016 | 66, F | Anemia, abdominal pain | Antrum | 11 | NA | EWSR1 gene rearrangement | No | No | DG | Yes | NA | NA |
| Hopp <i>et al</i> [4], 2019 | 24, M | Abdominal pain, nausea, vomiting | Posterior wall | 10 | keratin OSCAR/ AE1/AE3, CD99 | NA | NA | Yes | DG | NA | NA | NA |
| Ye <i>et al</i> [1], 2021 | 55, M | fatigue and fever, lose weight | posterior wall | 7.5 | CD99, CD57, CD56, vimentin | NA | No | No | DG + Roux-en-Y | No | 8 | Alive |

CG: Curative gastrectomy; DG: Distal gastrectomy; NA: Not available; RDG + Roux-en-Y: Distal gastrectomy + Roux-en-Y gastrojejunostomy; SG: Subtotal gastrectomy; TG: Total gastrectomy; TDPSLA: Total gastrectomy + distal pancreatectomy + splenectomy + left adrenalectomy; WR: Wedge resection.

primitive mesenchymal cells and possess limited neural differentiation potential[18,19]. This explains why they are typically located in the bone, peripheral nerves, and soft tissue[20].

EES grows slowly, and the mass diameter can exceed 20 cm; however, patients often do not exhibit any evident clinical symptoms[21]. Ye *et al*[1] summarized 12 cases of primary gastric ES and found that it tends to occur more often in women, has nonspecific clinical manifestations, and predominantly



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Figure 1 Abdominal contrast-enhanced ultrasound. A: Hepatic arterial phase; B: Hepatic portal vein phase; C: Hepatic delay phase.



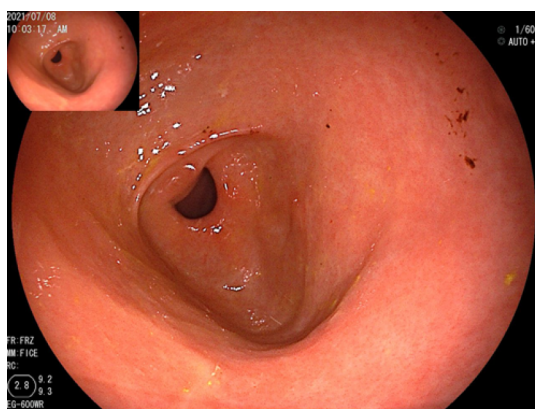
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Figure 2 Contrast-enhanced abdominal computerized tomography. A: Hepatic arterial phase; B: Hepatic portal phase; C: Hepatic venous phase.

presents as single tumors larger than 5 cm in diameter. Therefore, primary gastric ES is difficult to detect and diagnose early. In our patient, the clinical manifestations were nonspecific, and she was admitted to the hospital for further examination because she inadvertently palpated and identified an abdominal mass, which was eventually found to be an abdominal tumor.

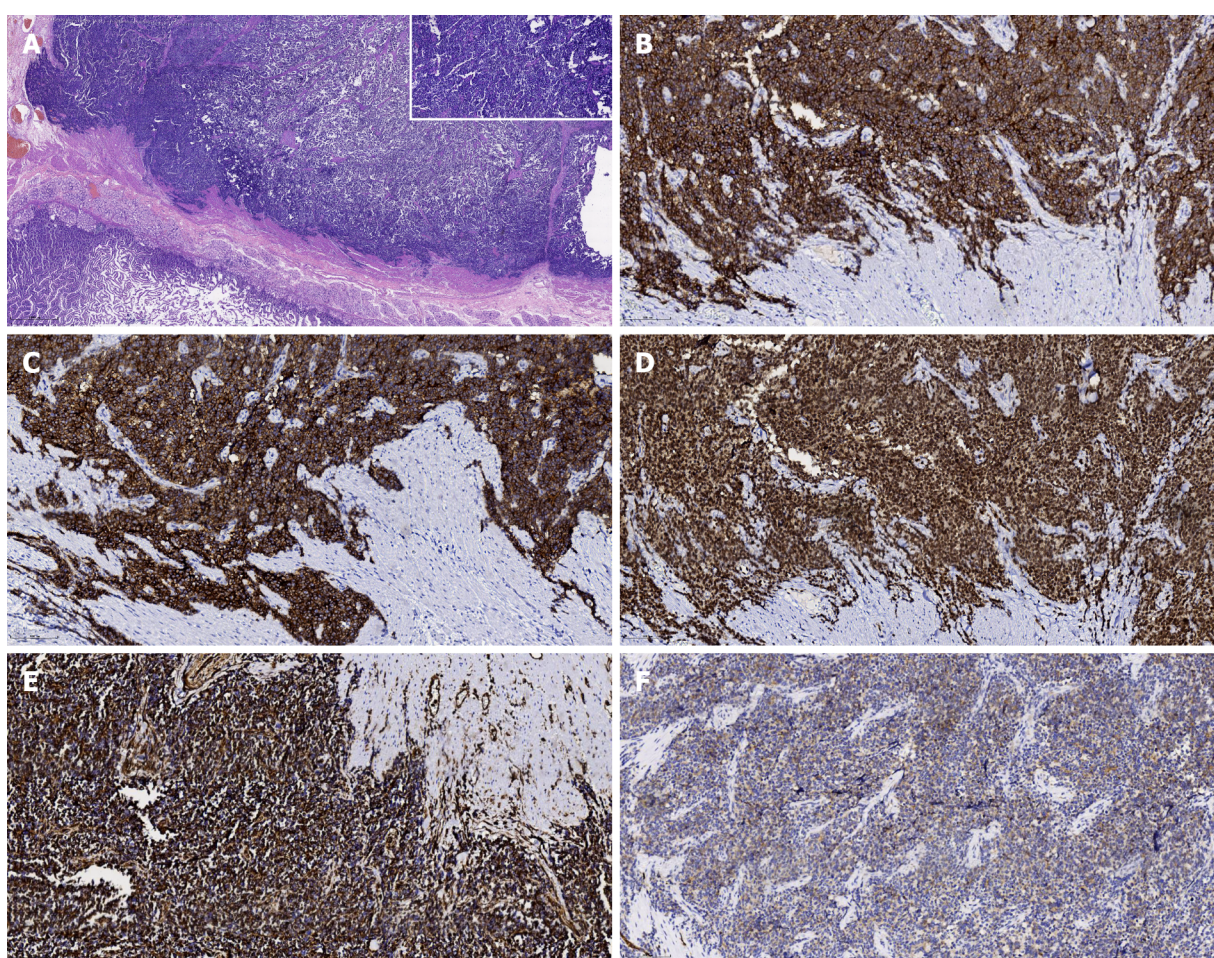
Clinical examination, imaging, and histology are nonspecific in evaluating rare tumors and distinguishing them from other solid masses. The imaging features of EES are nonspecific, with fluoro-18 fluorodeoxyglucose positron emission tomography/CT showing hypermetabolic lesions and CT/Magnetic resonance imaging typically showing an enhanced, bulky, soft-tissue lesion with internal heterogeneity if the tumor grows sufficiently large, secondary to ulceration, hemorrhage, and/or necrosis[1,4]. Calcification has been reported in up to 25% of cases[4]. On ultrasonography, EES usually appears as a low-echogenicity heterogeneous mass with intratumoral blood flow signals[22]. Owing to radiographic similarities, EES in abdominal and pelvic cavities may be frequently mistaken for gists[5], which was also the case in the preoperative imaging examination of this patient.

Pathological diagnosis is the gold standard for EES diagnosis. Histologically, ES presents with undifferentiated uniform small round cells, absence of nucleoli, a finely granular chromatin pattern, and a scanty cytoplasm with varying degrees of neuroectodermal differentiation[23]. Therefore, EES diagnosis relies on a constellation of immunohistochemical features as there is no pathognomonic marker, and these features include round cell morphology and characteristic immunohistochemistry findings, such as CD99, FLI-1, vimentin, NKX2, ERG, NSE, Leu-7, and SYN positivity[24,25].



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Figure 3 Esophagogastroduodenoscopy revealed that the gastric antrum mucosa was intact and smooth.



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Figure 4 Postoperative pathological examination. A: Hematoxylin and eosin staining. The tumor was identified to originate from the serous layer of the stomach and involve the muscularis externa of the stomach (20 ×). Upper right inset indicates small round cells with different sizes (original magnification, 200 ×); B-F: CD99-positive, vimentin-positive, FLI-1-positive, NSE-positive, and SYN-positive cells (immunohistochemistry staining, 200 ×).

CD99 is a single-chain type-1 glycoprotein that is highly sensitive but not specific, and its presence can be noted in a lot of other neoplastic conditions[23,24,26]. FLI-1 has recently been discovered as a DNA-binding transcription factor involved in t(11;22) translocation; although FLI-1 immunohistochemistry is typically positive in ES (about 75% of cases), it is also nonspecific and can occur in mucosal melanoma and neuroendocrine carcinoma[24,26]. Immunohistochemical studies may include an epithelial marker (pancytokeratin, such as CKAE1/AE3, epithelial membrane antigen, or OSCAR), a neuroendocrine marker (synaptophysin), a muscle marker (desmin, myogenin, or MYOD1), and S100



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Figure 5 Contrast-enhanced abdominal computerized tomography 11 mo postoperatively.

(to exclude melanoma)[26]. For patients suspected as having EES, fluorescence in situ hybridization is necessary to confirm the pathognomonic molecular abnormalities of t(11;22) (q24; q12)[24,25].

There is no unified standard for the treatment of EES. Currently, surgical resection and chemoradiotherapy are the most widely used treatments[25]. In our patient, the tumor lesion was relatively isolated and had not spread to the abdominopelvic cavity, thus allowing for its complete excision. ES is a radiosensitive tumor, and isolated radiotherapy can be used as a local treatment. Both ES and EES possess the EWSR1-ETS fusion protein and belong to the ES family. Therefore, EES is similarly sensitive to radiotherapy. When it is not possible to obtain wide margins because of the presence of fixed structures, such as vessels and/or nerves, postoperative radiotherapy can be implemented for better local control[27]. Accordingly, event-free survival at 5 years was better when surgery was followed by radiotherapy (70%) compared to only surgery (59%)[28]. However, our patient refused to undergo radiation therapy after radical surgery. Two months after the surgery, she underwent chemotherapy which comprised alternating cyclophosphamide, vincristine-doxorubicin, and ifosfamide-etoposide every 3 wk. At the time of the writing of this report, she is alive and shows no signs of recurrence 11 mo after the operation.

CONCLUSION

Taken together, primary gastric ES is difficult to diagnose early because of its rarity and nonspecific symptoms, signs, and imaging features. We believe this rare case is of importance for clinicians and radiologists to enhance their awareness of early identification and diagnosis of this type of tumor.

FOOTNOTES

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