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The *WJCC* is now indexed in Science Citation Index Expanded (also known as SciSearch®), Journal Citation Reports/Science Edition, Scopus, PubMed, and PubMed Central. The 2020 Edition of Journal Citation Reports® cites the 2019 impact factor (IF) for *WJCC* as 1.013; IF without journal self cites: 0.991; Ranking: 120 among 165 journals in medicine, general and internal; and Quartile category: Q3. The *WJCC*'s CiteScore for 2019 is 0.3 and Scopus CiteScore rank 2019: General Medicine is 394/529.

RESPONSIBLE EDITORS FOR THIS ISSUE

Production Editor: Ji-Hong Lin; Production Department Director: Xiang Li; Editorial Office Director: Jin-Lai Wang.

NAME OF JOURNAL

World Journal of Clinical Cases

ISSN

ISSN 2307-8960 (online)

LAUNCH DATE

April 16, 2013

FREQUENCY

Thrice Monthly

EDITORS-IN-CHIEF

Dennis A Bloomfield, Sandro Vento, Bao-Gan Peng

EDITORIAL BOARD MEMBERS

<https://www.wjgnet.com/2307-8960/editorialboard.htm>

PUBLICATION DATE

June 26, 2021

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INSTRUCTIONS TO AUTHORS

<https://www.wjgnet.com/bpg/gerinfo/204>

GUIDELINES FOR ETHICS DOCUMENTS

<https://www.wjgnet.com/bpg/GerInfo/287>

GUIDELINES FOR NON-NATIVE SPEAKERS OF ENGLISH

<https://www.wjgnet.com/bpg/gerinfo/240>

PUBLICATION ETHICS

<https://www.wjgnet.com/bpg/GerInfo/288>

PUBLICATION MISCONDUCT

<https://www.wjgnet.com/bpg/gerinfo/208>

ARTICLE PROCESSING CHARGE

<https://www.wjgnet.com/bpg/gerinfo/242>

STEPS FOR SUBMITTING MANUSCRIPTS

<https://www.wjgnet.com/bpg/GerInfo/239>

ONLINE SUBMISSION

<https://www.f6publishing.com>

Adult rhabdomyosarcoma originating in the temporal muscle, invading the skull and meninges: A case report

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Author contributions: Wang GH and Chu ZM designed the research; Shen J and Chu ZM performed the research; Shen HP analyzed the data; Wang GH and Shen HP contributed to manuscript drafting; all authors approved the submission of the final version.

Informed consent statement: Informed written consent was obtained from the patient for publication of this report and any accompanying images.

Conflict-of-interest statement: The authors have no conflicts of interest to declare.

CARE Checklist (2016) statement: The authors have read the CARE Checklist (2016), and the manuscript was prepared and revised according to the CARE Checklist (2016).

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Abstract

BACKGROUND

Rhabdomyosarcoma (RMS) is a rare malignant tumor of mesenchymal origin that mainly affects children. Spindle cell/sclerosing RMS (SSRMS) is even rarer. It is a new subtype that was added to the World Health Organization disease classification in 2013. To the best of our knowledge, this is the first reported case of adult SSRMS disease classification originating in the temporal muscle.

CASE SUMMARY

SSRMS originating in the temporal muscle of a male adult enlarged rapidly, destroyed the skull, and invaded the meninges. The tumor was completely removed, and the postoperative pathological diagnosis was SSRMS. Postoperative recovery was good and chemotherapy and radiotherapy were given after the operation. Followed up for 3 mo, no tumor recurred.

CONCLUSION

RMS is one of the differential diagnoses for head soft tissue tumors with short-term enlargement and skull infiltration. Preoperative computed tomography or magnetic resonance imaging is necessary for early detection of tumor invasion of the skull and brain tissue.

Key Words: Rhabdomyosarcoma; Temporal Muscle; Skull; Dura Mater; Adult; Case report

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Core Tip: Rhabdomyosarcoma (RMS) is a rare malignant tumor of mesenchymal origin

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Manuscript source: Unsolicited manuscript

Specialty type: Oncology

Country/Territory of origin: China

Peer-review report's scientific quality classification

Grade A (Excellent): 0
Grade B (Very good): 0
Grade C (Good): C
Grade D (Fair): 0
Grade E (Poor): 0

Received: February 8, 2021

Peer-review started: February 8, 2021

First decision: March 7, 2021

Revised: March 13, 2021

Accepted: May 7, 2021

Article in press: May 7, 2021

Published online: June 26, 2021

P-Reviewer: Gupta R

S-Editor: Gong ZM

L-Editor: Filipodia

P-Editor: Wang LL



that mainly affects children. Spindle cell/sclerosing RMS (SSRMS) is even rarer. We describe an adult case of SSRMS originating in the temporal muscle. The tumor rapidly enlarged, destroyed the skull, and invaded the meninges. The tumor was completely removed. The postoperative pathological diagnosis was SSRMS. This case report provides complete imaging data of tumor progression. To our knowledge, this case is the first reported adult SSRMS originating from the temporal muscle.

Citation: Wang GH, Shen HP, Chu ZM, Shen J. Adult rhabdomyosarcoma originating in the temporal muscle, invading the skull and meninges: A case report. *World J Clin Cases* 2021; 9(18): 4866-4872

URL: <https://www.wjgnet.com/2307-8960/full/v9/i18/4866.htm>

DOI: <https://dx.doi.org/10.12998/wjcc.v9.i18.4866>

INTRODUCTION

Rhabdomyosarcoma (RMS) is a rare, highly aggressive, rapidly growing mesenchymal malignancy that is more common in children[1]. Spindle cell/sclerosing RMS (SSRMS) is even rarer. It is a new subtype that was added to the disease classification of the World Health Organization (WHO) in 2013. RMS mainly occurs in the head and neck area[2]. It is very rare that RMS originates from the temporal muscle. We report a case of adult SSRMS that originated from the temporal muscle, destroyed the skull, and invaded the dura mater. As far as we know, there have been no previous reports of a case like this one.

CASE PRESENTATION

Chief complaints

A 55-year-old male patient was admitted to our hospital with a lump in the left temporal region.

History of present illness

Two months prior to admission, the patient noticed a lump in his left temporal scalp. The patient had no headaches or nausea and vomiting. The patient came to the outpatient department. A computed tomography (CT) scan was done and surgery was recommended, but the patient refused. The tumor grew slowly.

History of past illness

The patient had a 2-year history of hypertension.

Personal and family history

The patient had a 10-year history of smoking. He denied any family history.

Physical examination

Physical examination revealed a 6 cm × 7 cm hard, painless mass in the left temporal region. There was no redness or swelling on the surface of the mass.

Laboratory examinations

Laboratory examination, including liver and renal functions, blood counts, electrolytes, and coagulation function were normal. Serum tumor markers, HIV antibody, tuberculosis, and syphilis were negative.

Imaging examinations

The first CT showed a subcutaneous mass in the left temporal region (Figure 1). The second CT revealed that the mass was enlarged and the adjacent skull was destroyed (Figure 2). Magnetic resonance imaging (MRI) revealed a mass in the left temporal muscle, with obvious enhancement around the tumor, but no enhancement in the center of the tumor (Figure 3).

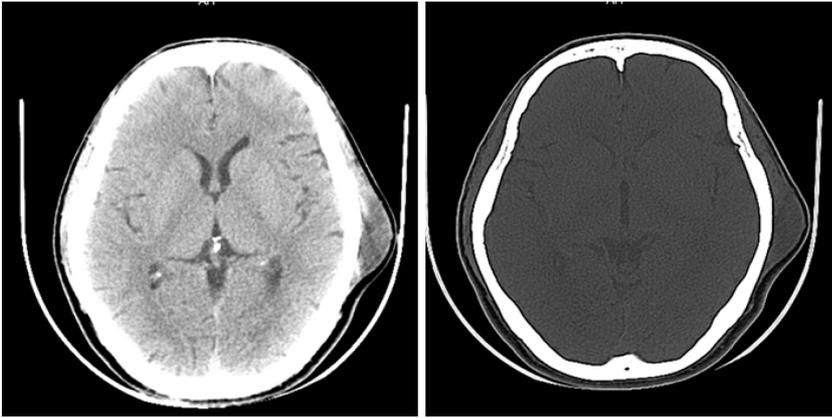


Figure 1 Computed tomography shows a subcutaneous mass in the left temporal region without skull destruction.

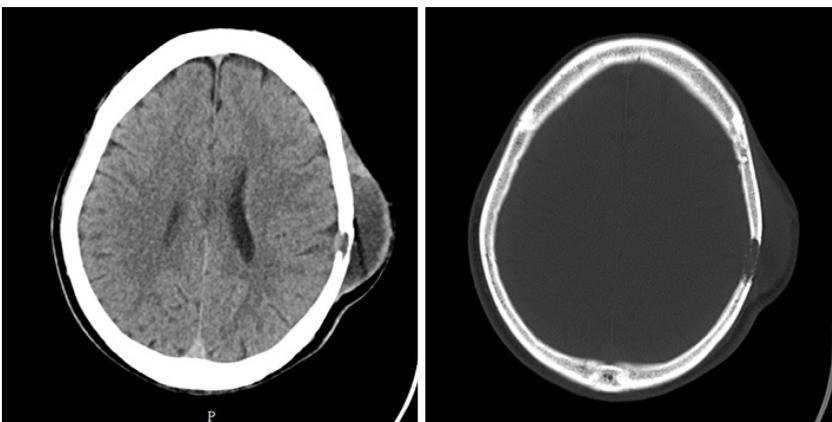


Figure 2 Computed tomography shows a soft tissue mass in the left temporal region and destruction of the adjacent skull.

FINAL DIAGNOSIS

The patient was admitted to our hospital with a diagnosis of left temporal soft tissue sarcoma, and metastatic tumor could not be ruled out. The pathology finding was SSRMS.

TREATMENT

The tumor was resected. During the operation, it was found that the tumor originated from the temporal muscle and had destroyed the skull and invaded the meninges. The tumor was completely removed with negative margins, and part of the skull and meninges were removed. Pathological evaluation of intraoperative frozen sections revealed malignant tumors, so cranioplasty was not performed. Chemotherapy and radiotherapy were given after the operation. Eight cycles of vincristine, ifosfamide, and etoposide were planned. A total of 50.4 Gy of radiation was administered.

OUTCOME AND FOLLOW-UP

The postoperative recovery was good. Tumor recurrence was not seen at the 3 mo follow-up. Pathological examination revealed that the tumor was composed of mildly atypical spindle cells arranged in a crossed bundle or spiral. A few rhabdomyoblasts were scattered among the spindle cells (Figure 4). Immunohistochemistry showed desmin (+), MyoD1 (+), Ki-67 (+) 60%, CD10 (+), SMA (-), GFAP (-), myosin (-), S-100 (-), GFAP (-), TLE (-), and s (-) cells (Figure 5). The diagnosis was SSRMS.

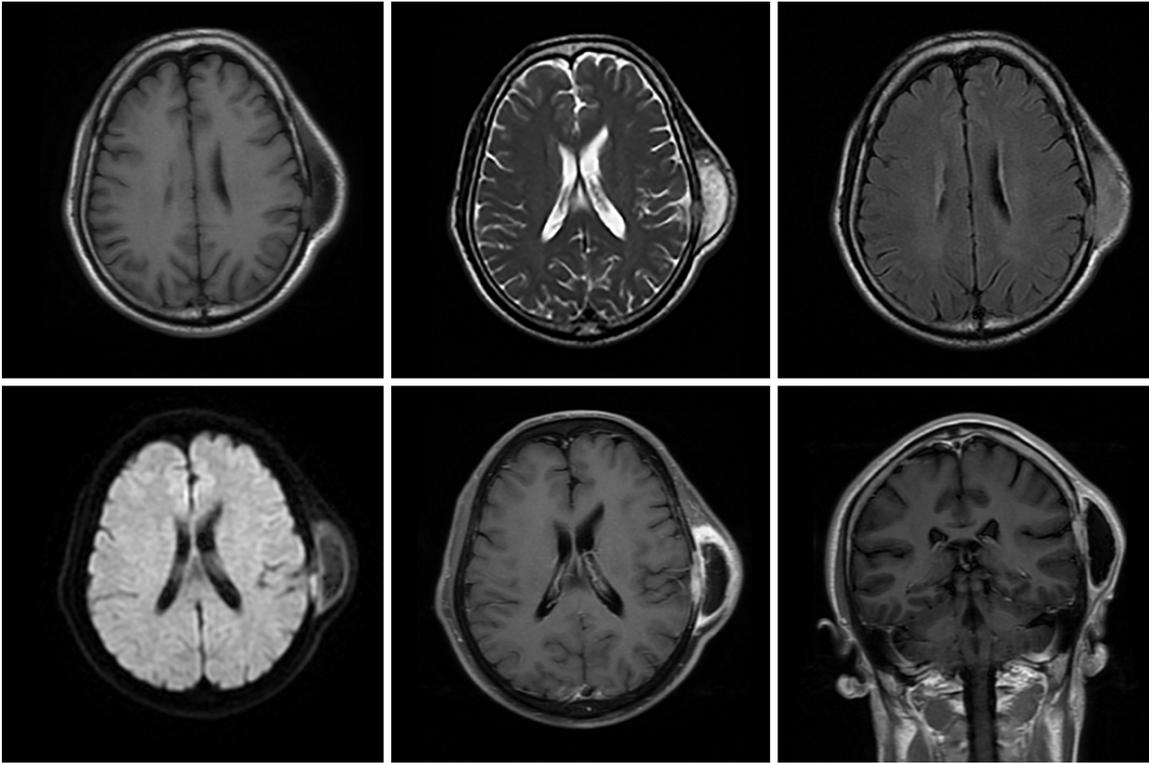


Figure 3 Magnetic resonance imaging shows a mass in the left temporal muscle, with obvious enhancement around the tumor, but no enhancement in the center of the tumor.

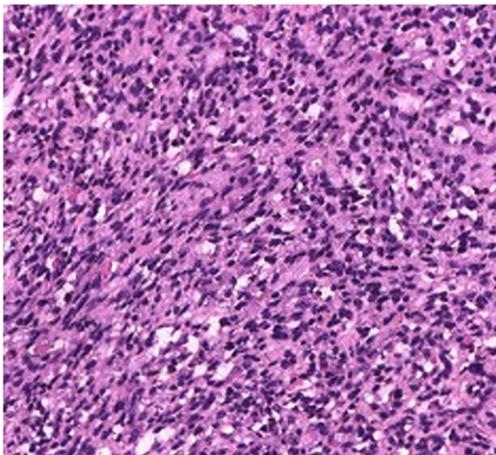


Figure 4 Pathological examination revealed a tumor composed of mildly atypical spindle cells arranged in a crossed bundle or spiral, and with a few rhabdomyoblasts scattered among the spindle cells (hematoxylin and eosin, $\times 200$).

DISCUSSION

Weber first reported RMS in 1854[3]. RMS is one of the most common tumors of children, about 250 new cases in children each year[1]. The age of onset has two peaks. The first is at 2 to 6 years of age and the second is at 14 to 18 years of age[2,4]. The median age is about 7 years[4]. Seventy percent of patients are younger than 10 years of age[5]. RMS has a very low incidence, and a high mortality rate, in adults[6].

RMS can originate from primitive mesenchymal cells anywhere in the body. Interestingly, most RMS tumors do not occur in muscles, but in areas where there is no muscle[2]. About 40% of RMSs occur in the head and neck region[2], followed by the urogenital tract, retroperitoneum, and limbs. The most common origins of head RMS are the orbits, nasopharynx, paranasal sinuses, middle ear, and external auditory canal [2,7]. The orbit is the most common single primary site[8]. This case originated from the temporal muscle, destroyed the skull, and invaded the meninges. Adult RMS is

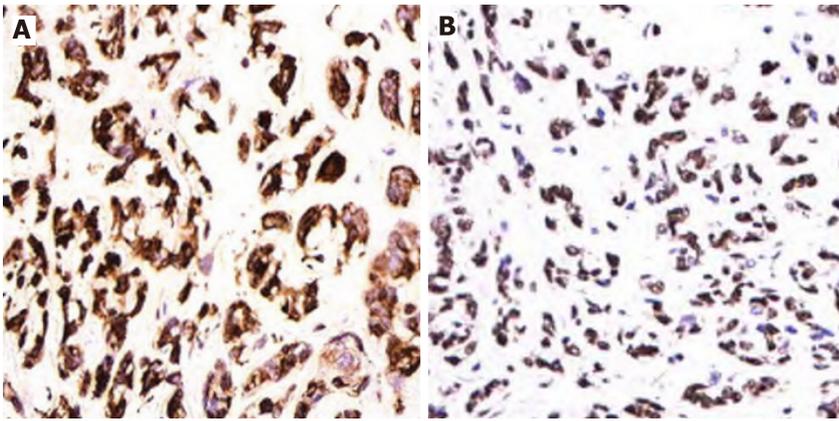


Figure 5 Immunohistochemical staining of tumor tissue. A: Desmin is strongly positive in the cytoplasm of tumor cells ($\times 200$); B: MyoD1 is strongly positive in the nucleus of tumor cells ($\times 200$).

very rare in clinical practice. As far as we know, no similar case has been reported in the literature.

Traditionally, three main RMS subtypes, embryonic, alveolar, and polymorphic, are recognized. However, other variants have been described, including SSRMS[9]. SSRMS was added to the WHO disease classification in 2013. This case was diagnosed as SSRMS based on pathological findings and immunohistochemistry. RMS mostly manifests as a rapidly increasing mass that can invade nearby tissues and metastasize to distant sites. The main clinical features of this case of RMS were a fast-growing painless temporal muscle mass, destruction of the skull, invasion of meninges, and normal skin. The imaging findings of SSRMS have definitive characteristics. In this case, enhanced MRI showed obvious enhancement around the tumor, but the tumor center had an extremely low signal, and no enhancement, similar to the characteristics described by Freling *et al*[10].

RMS usually requires a variety of treatment modalities, depending on the tumor location, size, and metastasis[11]. It is usually recommended to completely remove the tumor if surgery will not cause significant loss of function[12]. Survival is better if a definite negative surgical margin is achieved[13], but in many cases, the tumor cannot be completely removed, and only biopsy is possible. The prognosis of RMS is related to age, site of origin, tumor size, and metastasis[14,15]. Chemotherapy can shrink tumors and reduce large tumors that cannot be completely resected to the extent that they are easier to remove[11]. RMS can easily metastasize to the bone marrow, and some small tumors that cannot be detected by imaging examinations may have spread to other parts of the body, which is why chemotherapy is needed[16]. Positive margins after MRS surgery will result in a higher local failure rate[17]. Radiotherapy can reduce the local failure rate after MRS surgery. Studies found that in patients receiving radiotherapy, there was no correlation between positive margins and local recurrence [17].

The prognosis of RMS is significantly improved by more aggressive comprehensive treatment. The 5-year disease-free survival rates of early and late localized tumors are 81% [18] and 41% [19] respectively. Maurer *et al*[20] reported that the 5-year survival rates were 92% for orbital tumors, 81% FOR non-parameningeal tumors, and 69% FOR parameningeal tumors. If RMS invades the meninges, brain, and cranial nerves, the prognosis is extremely poor, often with rapid recurrence after surgical resection[21] and a median survival of 5-9 mon[22]. Among the reported cases, there was no 5-year survival[23,24]. In this case, chemotherapy and radiotherapy were still given even though although the tumor was completely resected because the tumor had invaded the meninges. Some authors recommend radical resection supplemented with radiation and chemotherapy[25].

CONCLUSION

Adult SSRMS originating in the soft tissues of the scalp is very rare. To our knowledge, this is the first reported case of SSRMS originating from the temporal muscle in an adult. It destroyed the skull and invaded the meninges. RMS is one of the differential diagnoses for head soft tissue tumors with short-term enlargement and skull infiltr-

ration. Preoperative CT or MRI is necessary for early detection of invasion of the skull and brain tissue.

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