

World Journal of *Clinical Cases*

World J Clin Cases 2021 June 26; 9(18): 4460-4880



OPINION REVIEW

- 4460** Surgery for pancreatic tumors in the midst of COVID-19 pandemic

Kato H, Asano Y, Arakawa S, Ito M, Kawabe N, Shimura M, Hayashi C, Ochi T, Yasuoka H, Higashiguchi T, Kondo Y, Nagata H, Horiguchi A

REVIEW

- 4467** Roles of exosomes in diagnosis and treatment of colorectal cancer

Umwali Y, Yue CB, Gabriel ANA, Zhang Y, Zhang X

MINIREVIEWS

- 4480** Dynamics of host immune responses to SARS-CoV-2

Taherkhani R, Taherkhani S, Farshadpour F

- 4491** Current treatment for hepatitis C virus/human immunodeficiency virus coinfection in adults

Laiwatthanapaisan R, Sirinawasatien A

- 4500** Anti-tumor effect of statin on pancreatic adenocarcinoma: From concept to precision medicine

Huang CT, Liang YJ

- 4506** Roles of vitamin A in the regulation of fatty acid synthesis

Yang FC, Xu F, Wang TN, Chen GX

ORIGINAL ARTICLE

Basic Study

- 4520** Identification of the circRNA-miRNA-mRNA regulatory network and its prognostic effect in colorectal cancer

Yin TF, Zhao DY, Zhou YC, Wang QQ, Yao SK

- 4542** Tetramethylpyrazine inhibits proliferation of colon cancer cells *in vitro*

Li H, Hou YX, Yang Y, He QQ, Gao TH, Zhao XF, Huo ZB, Chen SB, Liu DX

Case Control Study

- 4553** Significance of highly phosphorylated insulin-like growth factor binding protein-1 and cervical length for prediction of preterm delivery in twin pregnancies

Lan RH, Song J, Gong HM, Yang Y, Yang H, Zheng LM

Retrospective Cohort Study

- 4559** Expected outcomes and patients' selection before chemoembolization—"Six-and-Twelve or Pre-TACE-Predict" scores may help clinicians: Real-life French cohorts results

Adhoute X, Larrey E, Anty R, Chevallier P, Penaranda G, Tran A, Bronowicki JP, Raoul JL, Castellani P, Perrier H, Bayle O, Monnet O, Pol B, Bourliere M

Retrospective Study

- 4573** Application of intelligent algorithms in Down syndrome screening during second trimester pregnancy
Zhang HG, Jiang YT, Dai SD, Li L, Hu XN, Liu RZ
- 4585** Evaluation of a five-gene signature associated with stromal infiltration for diffuse large B-cell lymphoma
Nan YY, Zhang WJ, Huang DH, Li QY, Shi Y, Yang T, Liang XP, Xiao CY, Guo BL, Xiang Y
- 4599** Efficacy of combination of localized closure, ethacridine lactate dressing, and phototherapy in treatment of severe extravasation injuries: A case series
Lu YX, Wu Y, Liang PF, Wu RC, Tian LY, Mo HY
- 4607** Observation and measurement of applied anatomical features for thoracic intervertebral foramen puncture on computed tomography images
Wang R, Sun WW, Han Y, Fan XX, Pan XQ, Wang SC, Lu LJ
- 4617** Histological transformation of non-small cell lung cancer: Clinical analysis of nine cases
Jin CB, Yang L
- 4627** Diagnostic value of amygdala volume on structural magnetic resonance imaging in Alzheimer's disease
Wang DW, Ding SL, Bian XL, Zhou SY, Yang H, Wang P
- 4637** Comparison of ocular axis and corneal diameter between entropion and non-entropion eyes in children with congenital glaucoma
Wang Y, Hou ZJ, Wang HZ, Hu M, Li YX, Zhang Z

Observational Study

- 4644** Risk factors for postoperative delayed gastric emptying in ovarian cancer treated with cytoreductive surgery and hyperthermic intraperitoneal chemotherapy
Cui GX, Wang ZJ, Zhao J, Gong P, Zhao SH, Wang XX, Bai WP, Li Y
- 4654** Clinical characteristics, gastrointestinal manifestations and outcomes of COVID-19 patients in Iran; does the location matters?
Mokarram P, Dalivand MM, Pizuorno A, Aligolighasemabadi F, Sadeghdoust M, Sadeghdoust E, Aduli F, Oskrochi G, Brim H, Ashktorab H
- 4668** AWGS2019 vs EWGSOP2 for diagnosing sarcopenia to predict long-term prognosis in Chinese patients with gastric cancer after radical gastrectomy
Wu WY, Dong JJ, Huang XC, Chen ZJ, Chen XL, Dong QT, Bai YY

Prospective Study

- 4681** Clinical outcomes and 5-year follow-up results of keratosis pilaris treated by a high concentration of glycolic acid

Tian Y, Li XX, Zhang JJ, Yun Q, Zhang S, Yu JY, Feng XJ, Xia AT, Kang Y, Huang F, Wan F

Randomized Controlled Trial

- 4690** Tenofovir disoproxil fumarate in Chinese chronic hepatitis B patients: Results of a multicenter, double-blind, double-dummy, clinical trial at 96 weeks

Chen XF, Fan YN, Si CW, Yu YY, Shang J, Yu ZJ, Mao Q, Xie Q, Zhao W, Li J, Gao ZL, Wu SM, Tang H, Cheng J, Chen XY, Zhang WH, Wang H, Xu ZN, Wang L, Dai J, Xu JH

SYSTEMATIC REVIEWS

- 4700** Mesenteric ischemia in COVID-19 patients: A review of current literature

Kerawala AA, Das B, Solangi A

- 4709** Role of theories in school-based diabetes care interventions: A critical review

An RP, Li DY, Xiang XL

CASE REPORT

- 4721** Alport syndrome combined with lupus nephritis in a Chinese family: A case report

Liu HF, Li Q, Peng YQ

- 4728** Botulinum toxin injection for Cockayne syndrome with muscle spasticity over bilateral lower limbs: A case report

Hsu LC, Chiang PY, Lin WP, Guo YH, Hsieh PC, Kuan TS, Lien WC, Lin YC

- 4734** Meigs' syndrome caused by granulosa cell tumor accompanied with intrathoracic lesions: A case report

Wu XJ, Xia HB, Jia BL, Yan GW, Luo W, Zhao Y, Luo XB

- 4741** Primary mesonephric adenocarcinoma of the fallopian tube: A case report

Xie C, Shen YM, Chen QH, Bian C

- 4748** Pancreas-preserving duodenectomy for treatment of a duodenal papillary tumor: A case report

Wu B, Chen SY, Li Y, He Y, Wang XX, Yang XJ

- 4754** Pheochromocytoma with abdominal aortic aneurysm presenting as recurrent dyspnea, hemoptysis, and hypotension: A case report

Zhao HY, Zhao YZ, Jia YM, Mei X, Guo SB

- 4760** Minimally invasive removal of a deep-positioned cannulated screw from the femoral neck: A case report

Yang ZH, Hou FS, Yin YS, Zhao L, Liang X

- 4765** Splenic Kaposi's sarcoma in a human immunodeficiency virus-negative patient: A case report

Zhao CJ, Ma GZ, Wang YJ, Wang JH

- 4772** Neonatal syringocystadenoma papilliferum: A case report
Jiang HJ, Zhang Z, Zhang L, Pu YJ, Zhou N, Shu H
- 4778** Disappeared intralenticular foreign body: A case report
Xue C, Chen Y, Gao YL, Zhang N, Wang Y
- 4783** Femoral neck stress fractures after trampoline exercise: A case report
Nam DC, Hwang SC, Lee EC, Song MG, Yoo JI
- 4789** Collision carcinoma of the rectum involving neuroendocrine carcinoma and adenocarcinoma: A case report
Zhao X, Zhang G, Li CH
- 4797** Therapeutic effect of autologous concentrated growth factor on lower-extremity chronic refractory wounds: A case report
Liu P, Liu Y, Ke CN, Li WS, Liu YM, Xu S
- 4803** Cutaneous myiasis with eosinophilic pleural effusion: A case report
Fan T, Zhang Y, Lv Y, Chang J, Bauer BA, Yang J, Wang CW
- 4810** Severe hematuria due to vesical varices in a patient with portal hypertension: A case report
Wei ZJ, Zhu X, Yu HT, Liang ZJ, Gou X, Chen Y
- 4817** Rare coexistence of multiple manifestations secondary to thalamic hemorrhage: A case report
Yu QW, Ye TF, Qian WJ
- 4823** Anderson-Fabry disease presenting with atrial fibrillation as earlier sign in a young patient: A case report
Kim H, Kang MG, Park HW, Park JR, Hwang JY, Kim K
- 4829** Long-term response to avelumab and management of oligoprogression in Merkel cell carcinoma: A case report
Leão I, Marinho J, Costa T
- 4837** Central pontine myelinolysis mimicking glioma in diabetes: A case report
Shi XY, Cai MT, Shen H, Zhang JX
- 4844** Microscopic transduodenal excision of an ampullary adenoma: A case report and review of the literature
Zheng X, Sun QJ, Zhou B, Jin M, Yan S
- 4852** Growth hormone cocktail improves hepatopulmonary syndrome secondary to hypopituitarism: A case report
Ji W, Nie M, Mao JF, Zhang HB, Wang X, Wu XY
- 4859** Low symptomatic COVID-19 in an elderly patient with follicular lymphoma treated with rituximab-based immunotherapy: A case report
Łęcki S, Wyżgolik K, Nicze M, Georgiew-Nadziakiewicz S, Chudek J, Wdowiak K

- 4866** Adult rhabdomyosarcoma originating in the temporal muscle, invading the skull and meninges: A case report
Wang GH, Shen HP, Chu ZM, Shen J
- 4873** *Listeria monocytogenes* bacteremia in a centenarian and pathogen traceability: A case report
Zhang ZY, Zhang XA, Chen Q, Wang JY, Li Y, Wei ZY, Wang ZC

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

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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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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.

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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).

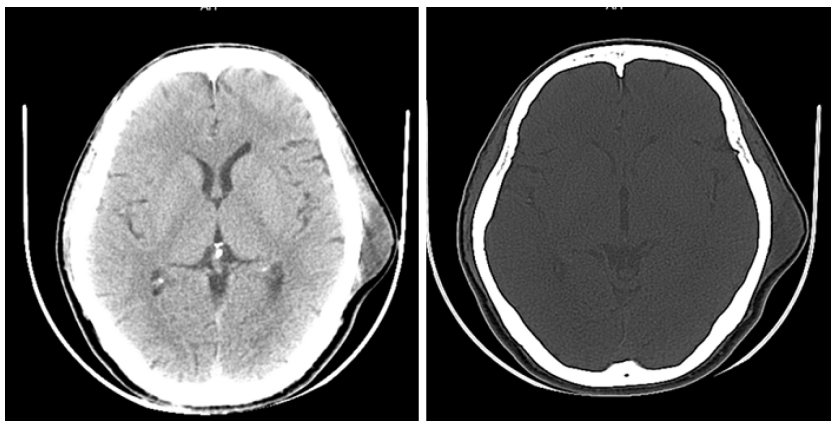


Figure 1 Computed tomography shows a subcutaneous mass in the left temporal region without skull destruction.

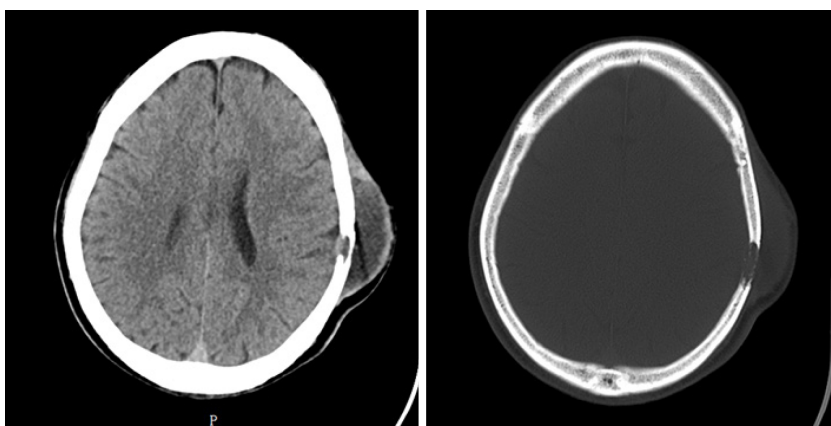


Figure 2 Computed tomography shows a soft tissue mass in the left temporal region and destruction of the adjacent skull.

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.

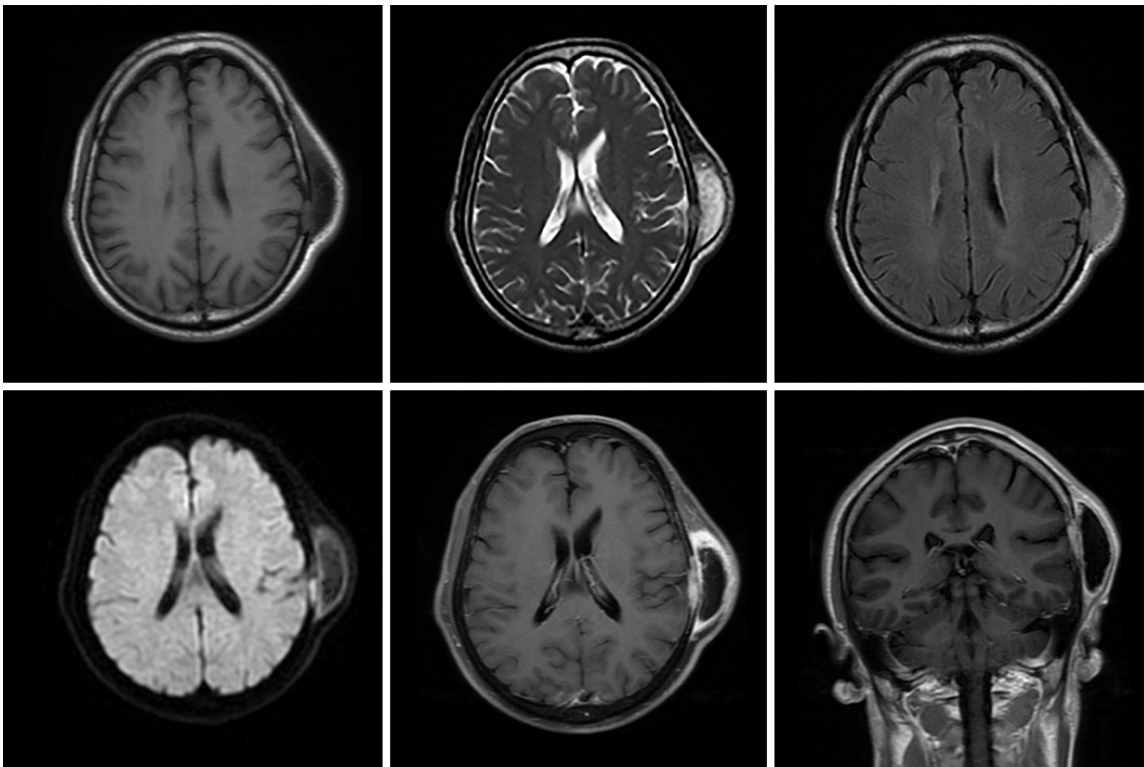


Figure 3 Magnetic resonance imaging shows a mass in the left temporal muscle, with obvious enhancement around the tumor, but no enhancement in the center of the tumor.

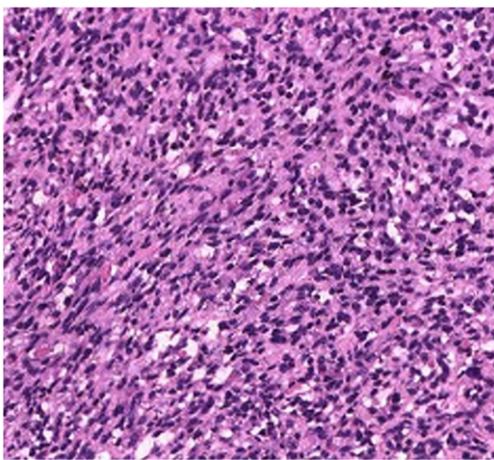


Figure 4 Pathological examination revealed a tumor composed of mildly atypical spindle cells arranged in a crossed bundle or spiral, and with a few rhabdomyoblasts scattered among the spindle cells (hematoxylin and eosin, $\times 200$).

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

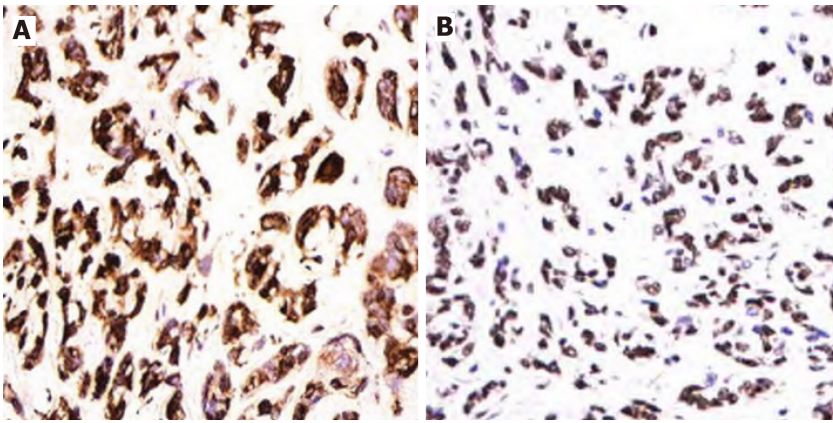


Figure 5 Immunohistochemical staining of tumor tissue. A: Desmin is strongly positive in the cytoplasm of tumor cells ($\times 200$); B: MyoD1 is strongly positive in the nucleus of tumor cells ($\times 200$).

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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