Pigmented villonodular synovitis of the temporomandibular joint: case report and the literature review for postoperative radiotherapy

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ABSTRACT

Pigmented villonodular synovitis (PVNS) is a benign proliferative disorder of the synovium that usually involves joints, tendon sheaths, and bursae. It presents rarely, however, in the temporomandibular joints (TMJs). This paper reports a 59-year-old female patient with PVNS of the TMJ and its clinico-pathologic features are discussed. The patient was treated with surgery and postoperative radiotherapy (PORT). Follow-up was conducted, and there were no recurrences, metastases, skin changes or joint stiffness noted. The main treatment of PVNS is surgical resection. However, postoperative radiotherapy is important for local control of extensive tumors or positive margins. We conducted a literature review for postoperative radiotherapy case reports related to PVNS of the TMJ.

CASE REPORT

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INTRODUCTION

Pigmented villonodular synovitis (PVNS) is a benign lesion of the synovium, affecting the joint spaces, tendon sheaths, and bursae. It usually presents in a monoarticular form and involves large joints or extremities [1]. PVNS of the temporomandibular joint (TMJ) is very rare.

The first documented case of PNVS was in 1852, and lesions of the flexor tendon sheath of the finger were reported by Chassaignac [2]. In 1941, Jaffe et al. established the pathology of PVNS [3]. We present a case of a 59-year-old female patient with PVNS of the right TMJ that initially presented as tinnitus and hearing loss. The role of radiotherapy in the treatment of diffuse PVNS of the TMJ is discussed in this report.

CASE REPORT

A 59 year-old Chinese female began to experience tinnitus in the right ear with hearing loss two-and-a-half years before presenting for treatment. A right temporal scalp mass was discovered three months before admission. Magnetic resonance images (MRI) displayed a heterogeneous soft tissue mass that abutted the right TMJ (Fig. 1). It also showed that the adjacent meninges were thickened, and that the surrounding bone and brain tissue were compressed and shifted. The infiltrative nature of the lesion was confirmed. Because of the destructive growth characteristics shown on the imaging, the main clinical differential diagnoses were of synovial sarcoma, idiopathic synovial osteochondromatosis, and rheumatoid arthritis (RA). A right middle cranial fossa intracranial resection was performed under general anesthesia. The right frontal valvular incision was taken to separate the

myocutaneous flap. Intraoperative observation showed the tumor of the right TMJ to be approximately 5cm×5cm×5cm in size. Postoperative pathology revealed the tumor to be diffuse PVNS with temporal bone tissue involvement (Fig. 2a). Histological examination showed that the mass contained multinucleated giant cells (Fig. 2b).

Because of the infiltrative nature of PVNS, PORT was recommended after resection. The postoperative changes of TMJ are shown in Fig 3. The lesion was surgically removed. Radiation therapy was carried out at 40 Gy in 20 fractions at 2.0 Gy per fraction. To reduce the dose to the parotid glands and adjacent brain tissue, we used the intensity modulated radiation therapy (IMRT) technique for the PVNS patient. In the later stages, during the IMRT treatment, this patient had grade 1/2 nausea, loss of appetite, and alopecia. There were no serious toxicities, such as dermatitis, malocclusion, blood toxicity, or xerostomia. Her tinnitus improved after treatment; however, her hearing problems did not improve. An MRI was performed every 3 months, with no evidence of recurrence one year after PORT. Radiotherapy did not cause local edema (Fig. 4). By the patient's 2 year follow-up, we found that she exhibited no facial paralysis, mandibular deviation, or malocclusion after post-operative radiotherapy. The maximum diameter of the mouth opening was 45 mm. Because of the rate of recurrence of this disease, continued long term follow-up is required. We will conduct long-term follow-up for this case.

DISCUSSION

Etiology & Demographics:

The etiology of PVNS is not clear, some researchers believe that the disease is an inflammatory reaction, and others think it is a tumor [3]. The incidence of PVNS is 2 to 8 per million people per year. There is no difference in the incidence between men and women. Patients are usually between 30 and 40 years old [4, 5]. The most common site of PVNS is the knee joint. PVNS of the TMJ is a highly rare disorder, with about 77 cases reported in the literature [6-10].

Clinical & Imaging findings:

PVNS is a benign lesion of the synovia, affecting the joint spaces, tendon sheaths, and bursae. It usually presents in a monoarticular form and involves large joints or extremities and causes discomfort and pain [1]. Two pathological types of PVNS are identified: the diffuse and the localized forms which are based on the involved area [11]. Histopathology of diffuse PVNS shows invasive pattern compared with localized PVNS. The diagnosis of PNVS is based on physical examination, imaging results and histological confirmation [3]. Pathological specimens can be characterized as villous, nodular, or villonodular, and in most cases visible hemosiderin deposits can be seen [12, 13]. The PVNS of TMJ can be presented as a pre-lump mass with locally destructive features. Due to its destructive and invasive properties, PVNS may involve the structure of the infraorbital fossae and the temporomandibular base [14]. CT scans can show the extent of hemosiderin, synovial lesions, and cystic changes and erosion of the bone. If there is extensive hemosiderin deposition, it shows an increase in density on CT. On T1 and T2 weighted MR images, hemosiderin shows either low or no signal. The most typical MRI feature of PVNS is a nodular mass with low intra-articular signal on T1, T2, and proton weighted images. Lesions and focal mass show the best on T2-weighted images, showing a low signal area. This is due to the deposition of hemosiderin. For localized PVNS, soft tissue masses with clear boundaries can be seen on MRI [15]. For diffuse PVNS, a mass with a border that is not very clear can be seen on MRI [12].

Treatment & Prognosis:

In terms of treatment, localized PVNS requires continuous follow-up or surgical resection. Usually surgical resection is the preferred treatment of localized PVNS. With the advancement of surgical techniques, arthroscopic resection has achieved good results in intra-articular masses [16]. Surgery is also considered to be the first treatment for diffuse PVNS. Currently, radiation therapy alone, or as an adjuvant therapy after synovectomy, has been studied. The treatment of diffuse PVNS should be individualized according to the specific location, the difficulty of local control and the recurrence rate of surgical treatment [3]. Because of some differing views about the treatments, especially PORT, we will conduct a detailed analysis. The treatment of most PVNS cases is exclusively primary surgical excision. Due to the characteristic of the synovial lesions and incomplete surgical resection, diffuse type cases of TMJ PVNS have a high rate of local recurrence. Radiation therapy after synovectomy has been studied by O'Sullivan et al. [17-22]. However, due to the possible toxicity of radiotherapy, Stephen R et al. [3] did not recommend postoperative adjuvant radiotherapy in PVNS cases.

We here report a diffuse TMJ PVNS case treated with PORT, with 2 years follow-up. To date, there has been no local recurrence in the patient. Because of its extreme aggressiveness, diffuse PVNS has a high local recurrence rate after surgery alone. Radiotherapy has been performed for PVNS patients with a high risk of recurrence in many institutions [17, 23-24]. In these studies, most of the patients failed the primary operation. Combined surgery with PORT has been performed in diffuse TMJ PVNS cases [25-27]. PORT is required when the following conditions are present: gross residual disease, positive margins, and extensive and invasive tumors. As the skull base and many normal tissues are closely linked, a complete resection may be difficult in the TMJ region. Diffuse PVNS, due to its nature, leads to irreversible bone and joint damage. Recurrence and repeated resection can aggravate the problem. As a way of local control, PORT provides a good therapeutic effect with minimal toxicity in diffuse PVNS of the TMJ. PORT is usually performed 3-4 weeks after surgery.

With the continuous advancement of radiotherapy, technology has developed from a traditional conventional treatment to three-dimensional conformal radiation therapy (3D-CRT), IMRT, image-guided radiation therapy (IGRT),

and other precise radiation therapy technologies. Radiotherapy allows precise positioning, accurate design, and precise treatment for patients. IMRT has the following advantages: it allows for the maximum dose for the tumor, offers protection of normal tissue by accurate tumor target location, and provides an evenly distributed target dose. Thus, IMRT technology delivers therapeutic radiation doses to the target tissues, while minimizing injury to surrounding normal tissue.

Although examples of PORT for extremity PVNS have been reported, the data available for the utility of PORT for PVNS of the TMJ is limited. In the study of this literature, acceptable doses are 35 Gy in 15 fractions, 40 Gy in 20 fractions, or 45 Gy in 25 fractions [6]. In our patient, the fractionation scheme was 40 Gy at 2.0 Gy/fraction daily for a total of 20 fractions. The radiation prescription volumes include the preoperative tumor volume and postoperative margin within half a centimeter.

Some researchers report concern for serious complications, such as skin necrosis and radiation-induced sarcoma [3]. Radiation-induced fractures and necrosis are unlikely to happen when the radiation dose is less than 50 Gy. Meanwhile, the radiation-induced secondary tumor risk is extremely low. The fractionation scheme administered to our patient was confirmed to be effective in other neoplasms, such as paragangliomas and schwannomas. O'Sullivan et al. reported a review of 14 patients with limb PVNS who received PORT [18]. After 12 months of follow-up, 12 individuals demonstrated local control of the disease, with no long-term side effects. Longer follow-up results by Griffin AM et al. showed that 41 patients with diffuse PVNS received PORT and had no complications [17]. Furthermore, 17 PVNS patients were treated with PORT by Horoschak et al. [19]. Followed for 46 months, most patients retained good function and no serious radiotherapy complications. Seven PVNS patients with PORT have been reported by Berger et al. [20]. All patients have been under control without long-term radiotherapy adverse events after 29 months of follow-up. With the development of scientific research and clinical progress, we will have a better understanding of the factors that determine whether toxicities occur, potential preventative therapies, and treatment strategies to minimize ionizing radiation exposure.

Differential Diagnoses:

1. Synovial sarcoma

On CT or MRI, synovial sarcoma is characterized by soft tissue around the joint. Bone erosion of synovial sarcoma is characterized by osteolytic destruction and there is no sclerotic zone of transition, no diffuse synovial thickening and no hemosiderin deposition. These imaging features are different from PVNS.

2. Idiopathic synovial osteochondromatosis

Diffuse PVNS must be distinguished from idiopathic synovial osteochondromatosis. Idiopathic synovial osteochondromatosis can demonstrate calcifications and ossifications on imaging.

3. Rheumatoid Arthritis (RA)

RA often invades multiple joints. PVNS usually manifests as a single joint of hemorrhagic arthritis. Single joint disease is a regularity of PVNS, but it can also rarely involve multiple joints.

4. Brown tumor

Brown tumor is one of the manifestations of hyperparathyroidism. Its most common clinical manifestations are painful symptoms in multiple parts of the body, difficulty walking, pathological fractures and limb deformities. Skeletal involvement often manifests as diffuse osteoporosis or multiple areas of bone resorption. Osteoporosis and incidental tumor-like changes are their primary imaging findings. The most characteristic finding of subperiosteal bone resorption is the most important and reliable X-ray sign for the diagnosis of brown tumor.

In summary, PVNS is, indeed, a rare lesion of the TMJ. PVNS of the TMJ requires primary surgical excision. PORT should be adopted in cases of incomplete surgical resection or extensive infiltrative PVNS. A suitable radiation dose is useful for treatment and does not cause serious complications. More cases must be analyzed in further studies. We also expect more and better treatments for PVNS of the TMJ in future.

TEACHING POINT

CT scans can show the extent of hemosiderin in pigmented villonodular synovitis (PVNS). If there is extensive hemosiderin deposition, it shows an increase in density on CT. On T1 and T2 weighted MR images, hemosiderin shows either low or no signal. The most typical MRI feature of PVNS is showing a low signal area on T2-weighted imaging, which is due to the deposition of hemosiderin. The main treatment of PVNS is surgical resection. However, postoperative radiotherapy is important for local control of extensive tumors or positive margins.

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FIGURES

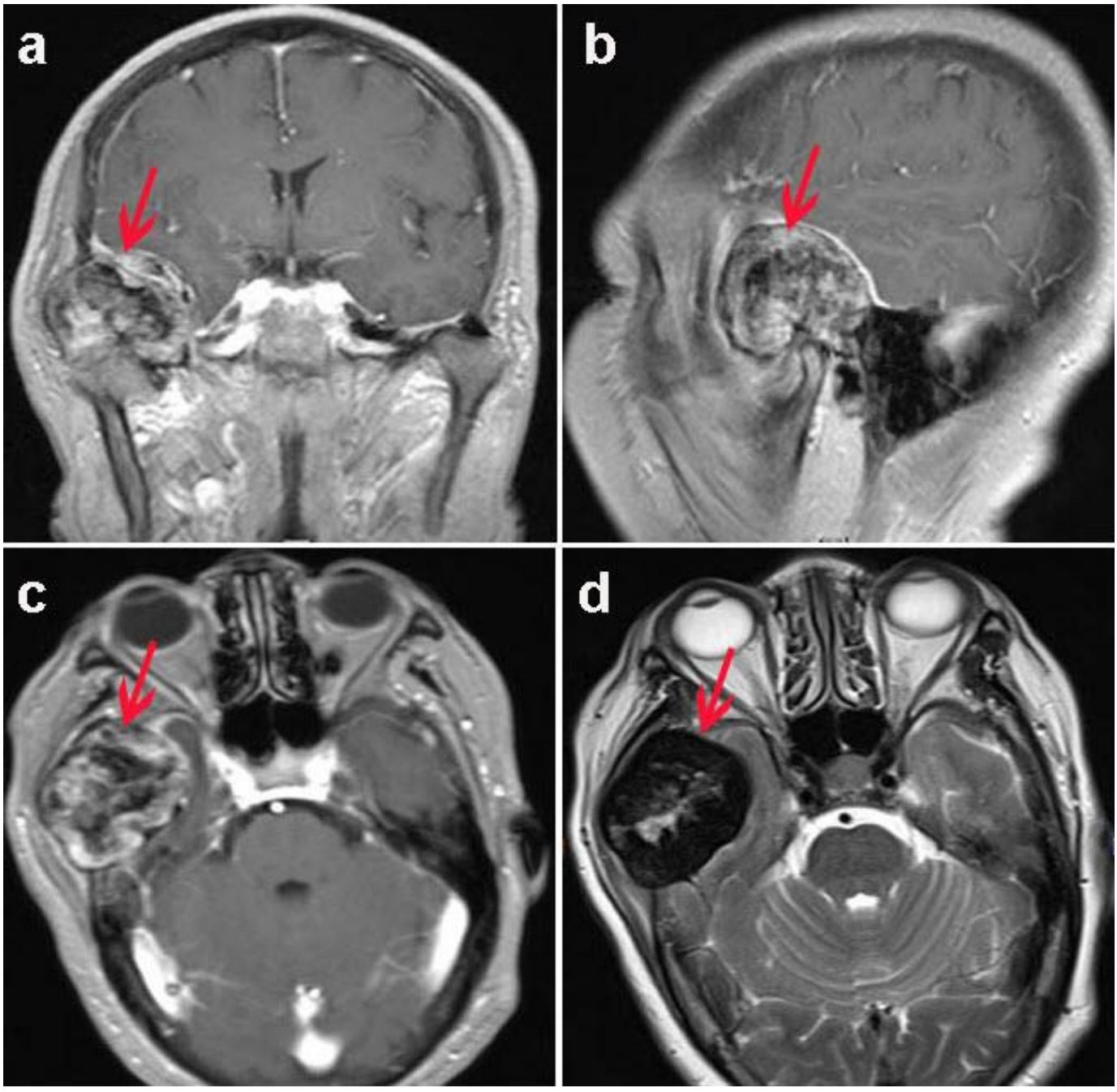

Figure 1: 59-year-old woman with PVNS of the right TMJ.

FINDINGS: T1-weighted sequences (a: coronal plane; b: sagittal plane; c: axial plane) and T2-weighted sequences (d: axial plane) show a large mass in the right TMJ (red arrows). An abnormal soft tissue mass presents in the right middle cranial fossae. The size is about 5.1×3.8×5.2 cm, which shows a low signal of T1WI, a low signal of T2WI, and a high signal at the center. After contrast administration, it shows heterogeneous enhancement, and the adjacent meninges are thickened with enhancement. Peripheral bone and brain tissue are displaced by mass effect. There is no ventricular enlargement. The brain sulcus is clear, no obvious widening, and there is no displacement of the midline structure.

TECHNIQUE: Philips Ingenia 1.5 T Magnetic Resonance System. Pre and post intravenous contrast administration (contrast agent used: Gadolinium-DTPA 0.2 ml/Kg). T1-weighted sequences (TR: 220, TE: 2.77). T2-weighted sequences (TR: 6000, TE: 95).

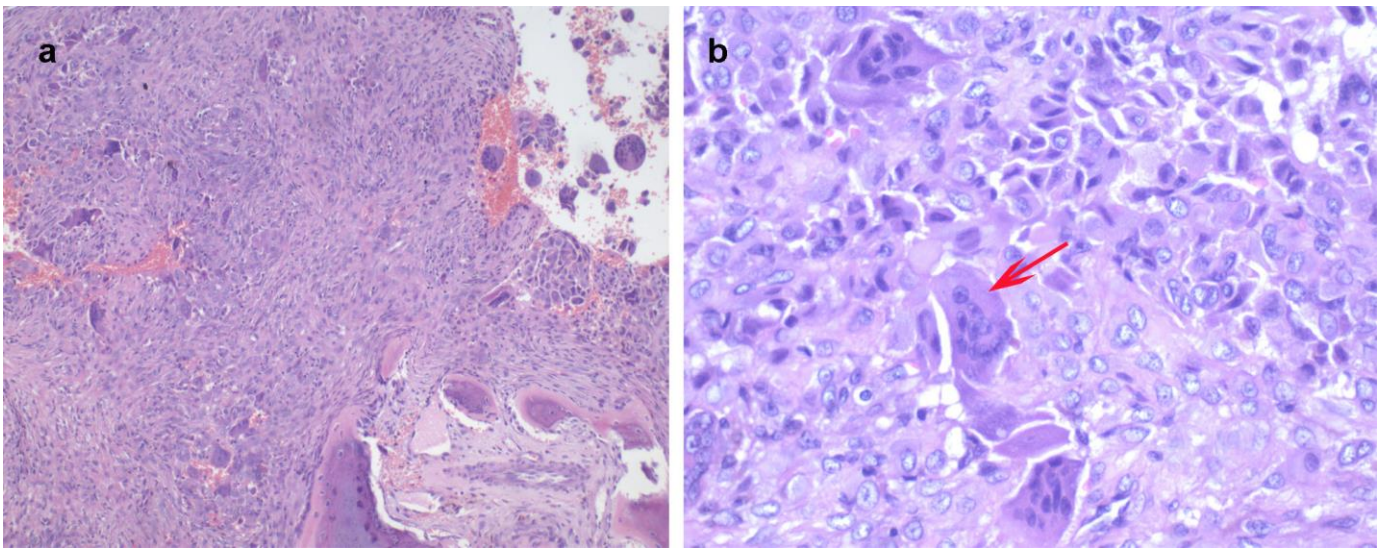

Figure 2: 59-year-old woman with PVNS of the right TMJ.

FINDINGS: (a) Postoperative pathology revealed the tumor to be diffuse PVNS with temporal bone tissue involvement. (b) Pathological features of PVNS include synovial hyperplasia with bands of cellular tissue containing multinucleated giant cells (red arrows).

TECHNIQUE: (a) hematoxylin-eosin stain, ×10 objective (b) hematoxylin-eosin stain, ×40 objective.

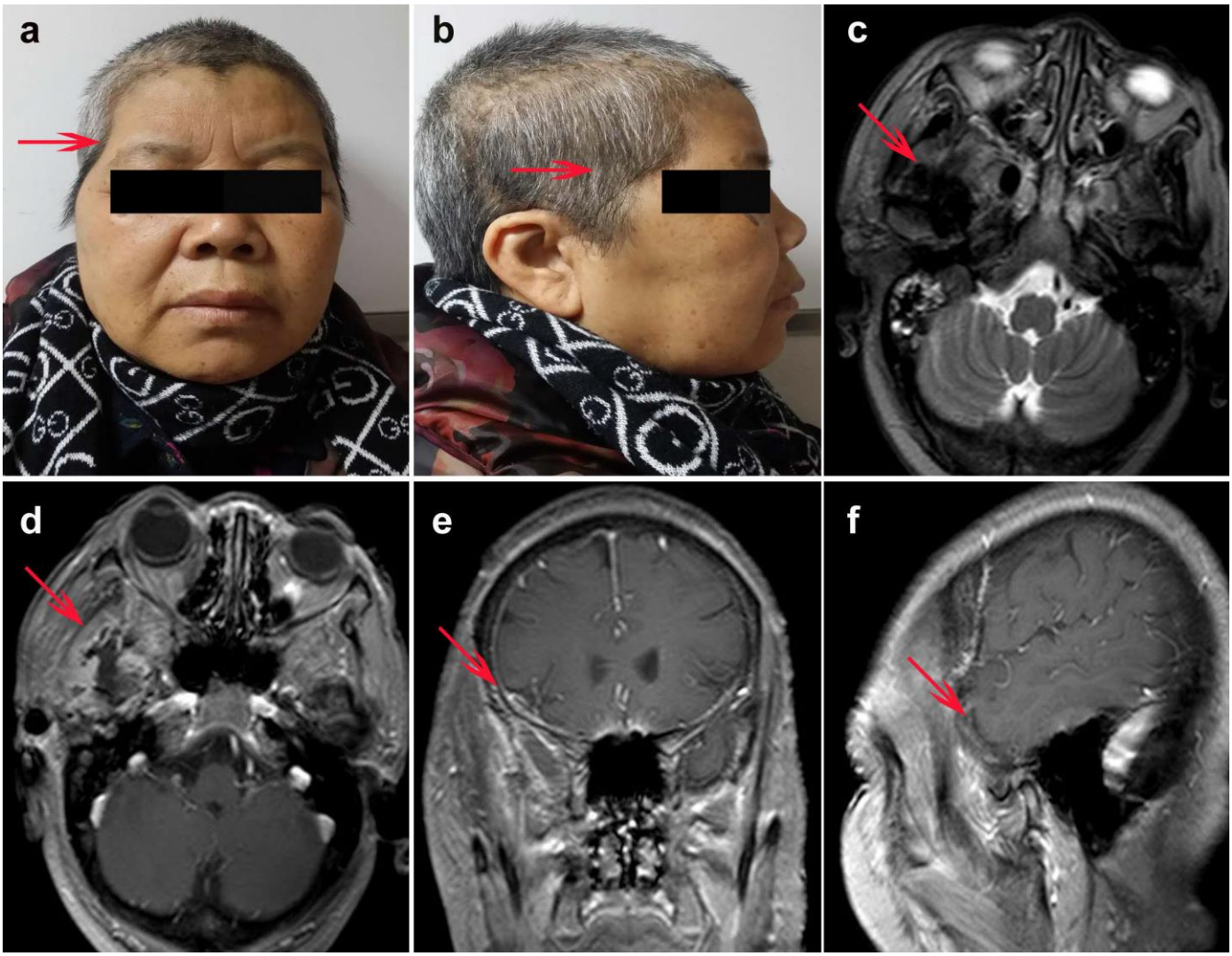

Figure 3: 59-year-old woman with PVNS of the right TMJ.

FINDINGS: Clinical photograph before radiation therapy (a and b). T2-weighted sequences (c: c: axial plane) and T1-weighted sequences (d: axial plane; e: coronal plane; f: sagittal plane) show postoperative changes of TMJ (as indicated by the red arrows). The right temporal lobe shows a low T1W and low T2W signal. The surrounding soft tissue is swollen and accumulated, and no obvious enhancement was noted after contrast administration.

TECHNIQUE: Philips Ingenia 1.5 T Magnetic Resonance System. Pre and post intravenous contrast administration (contrast agent used: Gadolinium-DTPA 0.2 ml/Kg). T1-weighted sequences (TR: 250, TE: 2.3). T2-weighted sequences (TR: 3508.41, TE: 80).

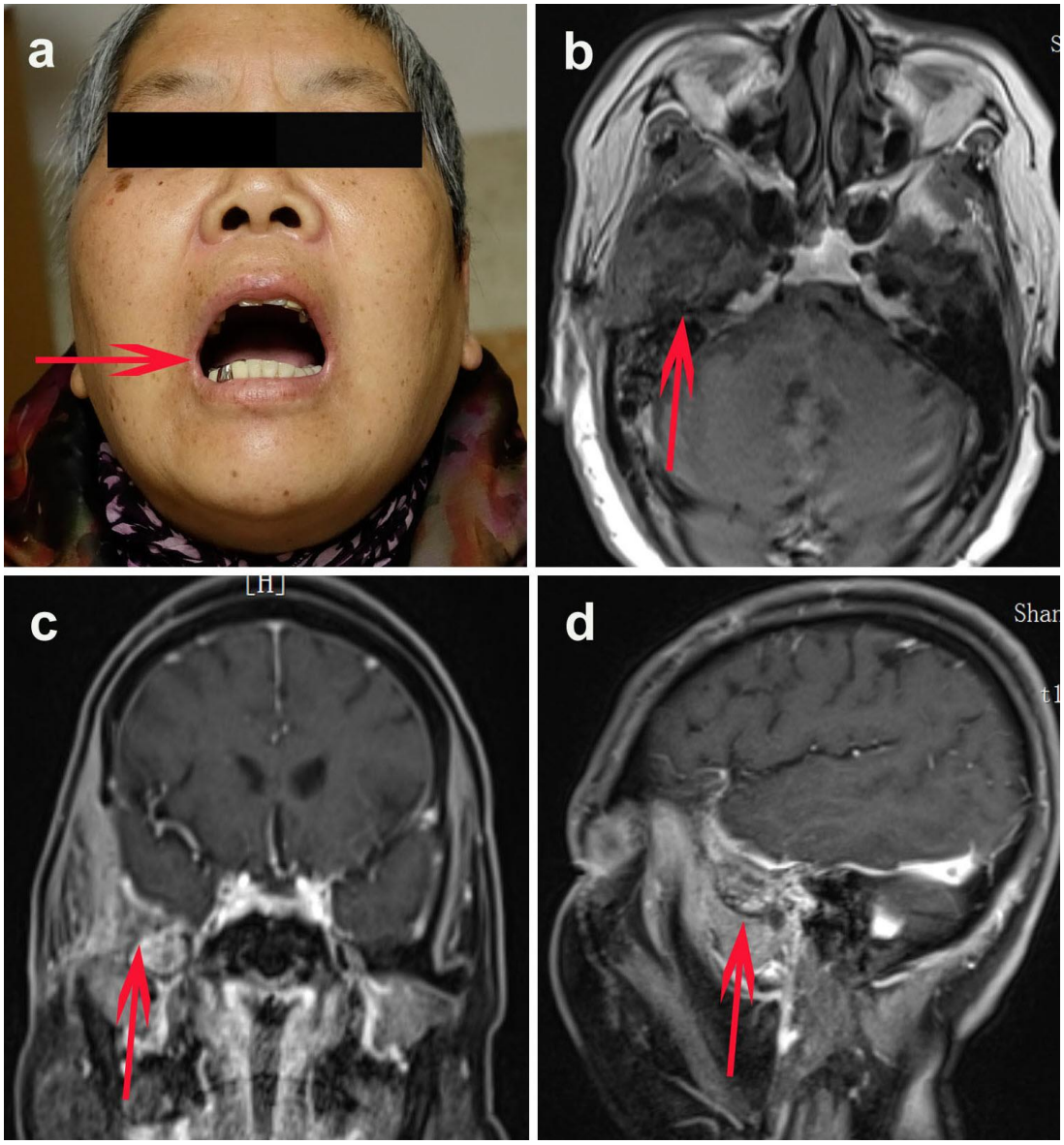

Figure 4: 59-year-old woman with PVNS of the right TMJ.

FINDINGS: Mandibular opening with minimal deviation (a). Follow-up MRI scan at 2 years demonstrates no recurrence on T1-weighted sequences (b: axial plane; c: coronal plane; d: sagittal plane) (as indicated by the red arrows). After the right axillary mass resection, the operation area is slightly disordered, and no obvious abnormal enhancement is observed after contrast administration.

TECHNIQUE: Philips Ingenia 1.5 T Magnetic Resonance System. T1-weighted sequences (TR: 220, TE: 2.77).

Etiology	Unknown
Incidence	2-8/1 million
Gender ratio	1:1
Age predilection	Between 30 and 40 years old
Risk factors	Unknown
Treatment	Surgical resection, radiotherapy
Prognosis	Often relapse
Findings on imaging	CT demonstrates the extent of hemosiderin. On T1 and T2 weighted MR images, hemosiderin presents either with low or no signal. PVNS is showing a low signal on T2-weighted imaging, showing a low signal area.

Table 1: Summary table for Pigmented villonodular synovitis (PVNS)

	Synovial sarcoma	Idiopathic synovial osteochondromatosis	Rheumatoid arthritis
X-Ray	Medium-density soft tissue mass with a round, elliptical or multinodular shape	Depending on the degree of calcification of the cartilage nodules, it may only be expressed as joint effusion, or multiple inferior osteochondral free bodies and overhanging body shadows in the joint, which are relatively uniform in size, and the typical osteochondral bodies are irregularly calcified. The high-density ring has a lighter-weight cancellous bone area at the center. Late manifestations of osteoarthritis, such as joint space narrowing, articular surface sclerosis, articular subchondral bone capsule changes.	It can be seen that the joints are swollen, the joint space is widened, the soft tissue around the joints is swollen, the osteoporosis and the articular cartilage are destroyed, but the subchondral cortical bone is intact, the local soft tissue is unclear, and the joint soft tissue can be seen to be swollen.
US	The appearance is not specific, mainly as a low echo mass.	The synovial sac can be seen with more anechoic areas, the capsule wall is thickened, the echo is increased, and nodular protrusions can be seen.	The joint capsule is thickened, and the echo of the joint exudate is homogeneous. The joint capsule is further thickened with a thickening of the synovial membrane, and the thickened synovial membrane is superficially echogenic as a hypoechoic or even anechoic region.
CT	Bone destruction and intragranular calcification	The center of the mass is light, with a ring-shaped calcium-like high density around it. Even the uncalcified cartilage CT can show a soft tissue density lower than the muscle.	Synovial cyst around the affected joint, cavity effusion, small concave defect at the end of the bone, or bone destruction in the bone, transverse or sagittal or coronal surface showing narrow joint space.
MRI - T1	The mass is higher, equal, and lower in intensity than muscle.	Intra-articular lobulated mass, T1 WI is equal or low signal compared to muscle.	moderate signals
MRI - T2	The lumps have a high signal and are clearly separated from the muscles.	Intra-articular lobulated masses, T2WI is high signal compared to muscle.	moderate signals
MRI - DWI	Multiple "pebble" nodules of similar size have low signal intervals and present a "paving stone" sign	Low signal intervals are seen in the mass.	markedly high signal
PET	Increased standard uptake value (SUV), high metabolism	SUV value does not increase, metabolism does not increase	Increased FDG uptake

Table 2: Differential diagnosis table for Pigmented villonodular synovitis (PVNS)

ABBREVIATIONS

IMRT = Intensity modulated radiation therapy
MRI = Magnetic resonance images
PORT = Postoperative radiotherapy
PVNS = Pigmented villonodular synovitis
RA = Rheumatoid arthritis
TMJ = Temporomandibular joint

KEYWORDS

case report; pigmented villonodular synovitis; PVNS; temporomandibular joint; TMJ; postoperative radiotherapy; PORT; prognosis

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