Imaging features of myopericytoma arising from the parotid gland
Report of 2 cases and literature review
Yao Pan, MDa, Lu Chen, MDb, Dan Shi, MDa, Ying Chen, MDa, Ri-Sheng Yu, MD, PhDa,*

Abstract
Rationale: Myopericytoma of the parotid gland is a rare condition of which preoperative definitive diagnosis is relatively challenging. The purpose of this case report is to highlight the radiologic features of myopericytoma of parotid gland.

Patient concerns: A 62-year-old man presented with a history of a walnut-size mass in left parotid gland when yawned for half-month, and a 48-year-old woman complaint about a grape-size, painless mass behind the right ear for a month.

Diagnoses: Radiological examinations suggested that both lesions were cyst-solid mixed lesions with relatively smoothed margins, with or without significant enhancement while the lesion without enhancement had a hemorrhage. Then a diagnosis of benign tumor arising from the parotid gland was made. Final diagnosis of myopericytoma was confirmed by histopathological and immunohistochemical examinations after surgical resection.

Interventions: Both patients underwent excision of the tumor and the superficial parotidectomy with facial nerve preservation.

Outcomes: Both patients recovered without any intraoperative or postoperative complication and had no signs of recurrence during a 17-month and 5-year follow-up.

Lessons: Parotid gland myopericytoma is an exceedingly rare tumor which diagnosis can be challenging, and this is the first published report specifying the magnetic resonance features of the disease.

Abbreviations: CD34 = the cluster of differentiation 34, CT = computed tomography, MRI = magnetic resonance imaging, SMA = smooth muscle actin.

Keywords: magnetic resonance imaging, myopericytoma, parotid gland

1. Introduction
The term myopericytoma was first proposed by Requena et al as an alternative designation for solitary myofibroma derived from myopericytes.[1] Myopericytoma describes a benign usually subcutaneous tumor that is composed of myoid-appearing oval to spindle-shaped cells with a striking tendency for concentric perivascular growth. The concept of perivascular myoid differentiation was established by Granter.[2] In 2002, the World Health Organization oficialized the term “myopericytoma” for use in clinical diagnoses.[3] Myopericytoma arises most commonly in middle adulthood, but may also occur in child.[4] Myopericytoma can be multifocal involving single or multiple anatomic regions,[1] and tends to occur predominantly in the skin and superficial soft tissue of the distal extremities (hand, foot, ankle, and leg), followed by the head and neck region, and the trunk.[6] Myopericytoma of the parotid gland is very rare. To date, only 5 published cases of myopericytoma arising from the parotid gland have been reported worldwide, and only 1 report described the computed tomography (CT) imaging features in detail. Current report provides a comprehensive description of parotid gland myopericytoma, including its complete clinical course and imaging findings. Reviews of past literature on this rare condition were also discussed.

1.1. Consent
The retrospective case report was approved by both patients, as well as the ethics committee of The Second Affiliated Hospital, Zhejiang University School of Medicine.

2. Case reports
2.1. Case one
A 62-year-old man was admitted to our hospital following the discovery of a walnut-size mass in left parotid gland when
yawned for half a month without any clinical symptoms. His medical history was otherwise unremarkable. Physical examination revealed a soft, painless, well circumscribed, and mobile mass in the left parotid region. Facial nerve function of the patient was normal on both sides. Ultrasound revealed an oval-shape, cyst-solid mixed, 3.0 cm × 2.0 cm mass located in the left parotid gland. Magnetic resonance imaging (MRI) showed a cyst-solid mixed, clearly defined mass in the left parotid gland. The solid component exhibits slightly high signal both on T1-weighted images and T2 weighted images, indicating the possibility of hemorrhage. The area of cystoid variation showed low signal with peripheral high signal on T1-weighted images and high signal on T2-weighted images (Fig. 1A and B). Gadolinium-enhanced MR imaging detected no enhancement (Fig. 1C).

Surgical excision of the superficial lobe of the left parotid gland and the tumor was performed. All facial nerve branches were preserved. The tumor was in the deep lobe and had invasion of adjacent tissue. The gross appearance of the resected specimen for the mass showed a soft, clear margined tumor of 2.3 cm × 2.0 cm × 3.0 cm in size and the content was black-brown. Photomicrograph of histology specimen showed numerous thin-walled vessels surrounded concentrically by proliferative spindle-shaped myoid tumor cells in peripheral zone (Fig. 2). Hemorrhage and necrosis in the nodule were evident. No malignant features such as nuclear anaplasia, increased mitoses, or infiltrative growth were found in the specimen. Immunohistochemical staining was positive for smooth muscle actin (SMA). The cluster of differentiation 34 (CD34) immunoperoxidase stain only decorated the endothelium of vessels, but the perivascular concentric myoid tumor cells were not immunoreactive. Based on these findings, this tumor was diagnosed as a benign myopericytoma arose from the parotid gland.

After 17 months of follow-up, the patient had no signs of recurrence.

2.1.1. Case two. A 48-year-old woman was referred to our hospital with primary complaint of a grape-sized painless mass behind the right ear for 1 month. Physical examination revealed a mass of 1 cm × 2 cm in size in the right parotid gland area. Ultrasoundography revealed an oval-shaped hypoecho lesion in the right parotid gland with clear border. Pre-contrast CT images demonstrated the presence of a 2.2 cm × 1.4 cm heterogeneous mass which was iso-density with central lower density and a relatively well-defined margin in the right parotid gland (Fig. 3A). After intravenous injection of contrast material, the mass demonstrated significantly heterogeneous enhancement with central irregular non-enhancement area (Fig. 3B). The CT values of the significant-enhanced areas in pre-contrast and enhanced scans were 44.82 HU and 157.32 HU, respectively. The lesion exhibited a solid iso-signal nodule on T1-weighted images (Fig. 4A), and high signal with central spotty higher signal on T2-weighted fat-suppression images (Fig. 4B). Enhanced images

Figure 1. A 62-yr-old man with myopericytoma in the left parotid gland. (A and B) T1-weighted images (A) and T2-weighted images (B) revealing a well defined, cyst-solid mixed mass with slightly high signal intensity in the solid portion; (C) Axial enhanced scan revealing no enhancement of the lesion. The areas of necrosis and cystic degeneration were non-enhanced.

Figure 2. Photomicrograph of histology specimen showed numerous thin-walled vessels surrounded concentrically by proliferative spindle-shaped myoid tumor cells in peripheral zone with obviously hemorrhage and necrosis. (Hematoxylin-eosin stain, original magnification, 100 × )
revealed obvious enhancement with mottled non-enhancement area (Fig. 4C).

Excision of the tumor and the superficial parotidectomy with facial nerve preservation were performed. The tumor was between the deep lobe and the superficial lobe with invasion of adjacent tissue. Histopathologic examination showed a proliferation of overlapping plump, spindle-shaped myoid cells in a concentric arrangement, intimately associated with thin-walled vascular channels (Fig. 5). The lesion cells showed positive immunohistochemical reactivity for SMA. Immunostaining was negative for CD34, CD31, desmin, and S-100. The pathologic diagnosis of the surgery specimen was myopericytoma.

The patient had no signs of recurrence during a 5-year follow-up.

3. Discussion
Myopericytoma is an uncommon tumor, and reports of myopericytoma in parotid gland are even more rare. Therefore, it is difficult for the surgeons and radiologists to make the accurate diagnosis. Benign myopericytoma is generally considered a slow-growing mass without pain. Myopericytoma usually follows a benign clinical course with local recurrence potential, while malignant myopericytoma has also been reported.[2–4] Histological findings of myopericytoma are characterized by the presence of round or oval-shaped cells with eosinophilic cytoplasm arranged circumferentially around vascular lumina in a multilayered pattern (onion skin), often benign with absence of mitoses, pleomorphism, or necrosis. It is believed that the myopericytoma cell of origin is either the pericyte or myofibro-

Figure 3. A 48-yr-old woman with myopericytoma in the right parotid gland. (A) Pre-contrast CT image revealing a 2.2 cm × 1.4 cm, relatively well defined, heterogeneous iso-density mass with central lower density in the right parotid gland; (B) Enhanced scan revealing mass with significantly heterogeneous enhancement and central irregular non-enhanced area. The CT values of the significant-enhanced areas in pre-contrast and enhanced scans were 44.82 HU and 157.32 HU, respectively.

Figure 4. (A and B) The lesion exhibited a solid iso-intensity nodule on T1-weighted images, and high signal intensity with central spot higher signal intensity on T2-weighted fat-suppression images; (C) Axial enhanced images revealing lesion with obvious enhancement and mottled non-enhancement area.
myopericytoma in the parotid gland. Kuczkowski et al reported a case of myopericytoma in the parotid gland showed 2 foci in the left parotid gland with significant heterogeneous enhancement.[13] Myopericytoma arising from other regions also most often appear as an enhanced T1 hypointense, T2 hyperintense mass.[10] The second case in present report had a significant enhancement with mottled non-enhancement area as well. We speculated that the obvious enhancement with or without hemorrhage and necrosis may be the imaging feature for myopericytoma of parotid gland. However, the first case in present report was not enhanced, which is quite different from previous findings. We suppose that this may be related to the hemorrhage in the lesion. To the best of our knowledge, the present report is the first to report hemorrhage in parotid gland myopericytoma. Thus, when a parotid gland mass with a well-defined cyst-solid imaging features and hemorrhage was detected, the diagnosis of myopericytoma should be considered in the differential diagnosis. However, future studies are still needed to verify this guess.

The common MRI features of these 2 cases were cyst-solid mixed appearance, and relatively smoothed margins with hyperintense on T2-weighted image in the solid component. We are the first to provide a comprehensive radiological description of parotid gland myopericytoma, including CT and MRI findings.

The differential diagnosis should include tumors with similar MR findings that are commonly seen in the parotid gland. Warthin’s tumors usually demonstrated a rounded or lobulated lesion with intermediate signal on both T1-weighted images and T2-weighted images, and focal T2-hyperintense area corresponding to the cystic components. However, the tumor usually shows rapid contrast enhancement and washout on post-contrast T1-weighted images.[13] The characteristics of pleomorphic adenoma are hyperintense areas on T2-weighted images with marked enhancement, fibrous capsules, and a lobulated contour. Cystic change and hemorrhage are often seen in larger tumors (>3 cm).[16] Basal cell adenoma shows relatively low signal intensity on T1-weighted images and hypointense to slightly hyperintense on T2-weighted images, with rapid and prolonged enhancement on dynamic scans. Basal cell adenoma sometimes has cystic or hemorrhagic components.[17] Mucoepidermoid carcinoma may be predominantly cystic or mixed cystic with solid mural components. The MRI findings have a tendency to be related to the histological grade.[18] There are some general features suggesting malignancy, such as irregular margins, extra-glandular infiltration, perineural spread, and secondary lymphadenopathies.[15]

Myopericytoma usually follows a benign clinical course, in our case, after 17-month and 5-year follow-up, both cases had no

### Table 1

| Lesion   | Age (yr) | Sex  | Location               | Maximum diameters | Surgical treatment                        | Follow-up | Reference |
|----------|----------|------|------------------------|-------------------|------------------------------------------|-----------|-----------|
| 1        | 40       | Female | Left parotid gland     | 2.5 cm            | Excision of the tumor and superficial parotidectomy | NS        | [9]       |
| 2        | 43       | Female | Right parotid gland    | 5.6 cm            | Excision of the tumor and total parotidectomy | R         | [11,12]  |
| 3        | 65       | Male  | Left parotid gland     | 1.4 cm            | Excision of the tumor and total parotidectomy | 2 yr, NR  | [13]      |
| 4        | 65       | Male  | Left parotid gland     | 1.2 cm            | Excision of the tumor and total parotidectomy | 2 yr, NR  | [13]      |
| 5        | 66       | Male  | Right parotid gland    | 0.9 cm            | Extracapsular dissection                   | 18 mo, NR | [14]      |
| 6        | 62       | Male  | Left parotid gland     | 3.0 cm            | Excision of the tumor and superficial parotidectomy | 17 mo, NR | Present case |
| 7        | 48       | Female | Right parotid gland    | 2.2 cm            | Excision of the tumor and superficial parotidectomy | 5 yr, NR | Present case |

NR = no recurrence, NS = not sure, R = recurrence.
signs of recurrence, but local recurrence and malignant myopericytoma have also been reported.\textsuperscript{[2–4]} It is generally accepted that parotid gland myopericytoma should be treated with wide surgical excision to prevent local recurrence. The superficial parotidectomy with facial nerve preservation is known as a widely acceptable procedure. Meanwhile, previous study suggested that via extracapsular dissection may provide improved functional and facial aesthetic outcomes for parotid neoplasms compared with superficial parotidectomy.\textsuperscript{[14]} Radiation has been used in selective cases to decrease the likelihood of recurrence, especially if there has been incomplete tumor excision.\textsuperscript{[15]} Ongoing molecular studies may offer more suitable options for patients with malignant myopericytoma in the future.

4. Conclusion

To the best of our knowledge, this is the first report to specify the MRI features of myopericytoma involving the parotid gland. Myopericytoma should be considered during the differential diagnosis of painless, cyst-solid mixed lesions with relatively smooth margins, high signal intensity on T2-weighted images, and obvious enhancement tumor involving parotid gland. It is worth noting that the tumor may present unenhanced mass due to hemorrhage sometimes.

Acknowledgments

We thank Xiao-Pei Xu for her excellent help in the preparation of the manuscript.

Author contributions

Conceptualization: Yao Pan, Lu Chen, Ri-Sheng Yu.
Data curation: Yao Pan, Lu Chen, Dan Shi.
Formal analysis: Dan Shi, Ying Chen.
Methodology: Yao Pan, Lu Chen, Ri-Sheng Yu.
Resources: Yao Pan, Lu Chen, Dan Shi, Ying Chen, Ri-Sheng Yu.
Supervision: Ying Chen, Ri-Sheng Yu.
Writing – original draft: Yao Pan, Lu Chen.
Writing – review & editing: Ying Chen, Ri-Sheng Yu.

References

[1] Requena L, Kutzner H, Hägel H, et al. Cutaneous adult myofibroma: a vascular neoplasm. J Cutan Pathol 1996;23:445–57.
[2] Granter SR, Radizadegan K, Fletcher CD. Myofibromatosis in adults, glomangiopericytoma, and myopericytoma: a spectrum of tumors showing perivascular myoid differentiation. Am J Surg Pathol 1998;22:513–25.
[3] McMenamin ME, Fletcher CDM. Malignant myopericytoma: expanding the spectrum of tumours with myopericytic differentiation. Histopathology 2002;41:450–60.
[4] Yaman H, Gerek M, Tosun F, et al. Myoepithelioma of the parotid gland in a child: a case report. J Pediatric Surg 2010;45:E5–7.
[5] Mentzel T, Dei Tos AP, Sapi Z, et al. Myopericytoma of skin and soft tissues: clinicopathologic and immunohistochemical study of 34 cases. Am J Surg Pathol 2006;30:104–13.
[6] Laga AC, Tajirian AL, Islam MN, et al. Myopericytoma: report of two cases associated with trauma. J Cutan Pathol 2008;35:866–70.
[7] Dray MS, McCarthy SW, Palmer AA, et al. Myopericytoma: a unifying term for a spectrum of tumours that show overlapping features with myofibroma. A review of 14 cases. J Clin Pathol 2006;59:67–73.
[8] Thompson LDR, Miettinen M, Weng BM. Sinonasal-type hemangio-pericytoma: a clinicopathologic and immunophenotypic analysis of 104 cases showing perivascular myoid differentiation. Am J Surg Pathol 2003;27:737–49.
[9] Jung Y-I, Chung Y-K, Chung S. Multiple myopericytoma of the face and parotid gland. Arch Plast Surg 2012;39:138–61.
[10] Yang JC, Ventescher AS, Koch MJ, et al. Myopericytoma at the cranio-cervical junction: clinicopathological report and review of a rare perivascular neoplasm. Neurosurgery 2019;85:E360–5.
[11] Xia L, Chen Y, Geng N, et al. Multifocal myopericytoma in the maxillofacial region: a case report. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2010;109:e59–62.
[12] Chu ZG, Yu JQ, Yang ZG, et al. Myopericytoma involving the parotid gland as depicted on multidetector CT. Korean J Radiol 2009;10:398–401.
[13] Kocialkowski J, Raepko R, Szwarska E. Myopericytoma of the parotid gland – a pathological conundrum. J Cranio-maxillo-facial Surg: official publication of the European Association for Cranio-Maxillo-Facial Surgery 2010;38:595–6.
[14] Bates AS, Craig P, Knepil GJ. Myopericytoma of the parotid region treated by extracapsular dissection. BMJ Case Rep 2014;2014:bcr2013201924.
[15] Cantissani V, David E, Sidiou PS, et al. Parotid gland lesions: multiparametric ultrasound and MRI features. Ultraschall Med (Stuttgart, Germany: 1980) 2016;37:454–71.
[16] Kato H, Kanematsu M, Minuta K, et al. Imaging findings of parapharyngeal space pleomorphic adenoma in comparison with parotid gland pleomorphic adenoma. Jpn J Radiol 2013;31:724–30.
[17] Mukai H, Motoori K, Horikoshi T, et al. Basal cell adenoma of the parotid gland; MR features and differentiation from pleomorphic adenoma. Dentomaxillofac Radiol 2016;45:20150322.
[18] Kashiwagi N, Dote K, Kawano K, et al. MRI findings of mucoepidermoid carcinoma of the parotid gland: correlation with pathological features. Br J Radiol 2012;85:709–13.
[19] Agrawal N, Nag K. Myopericytoma of the thoracic spine: a case report and review of literature. Spine J 2013;13:e23–7.