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Tumor Discovery

CASE REPORT

Odontogenic myxofibroma arising in the mandibular angle of a child with long-term follow-up: A case report

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Abstract

Odontogenic myxofibroma of the jawbone is a rarity in children, and there are few reports of cases with long-term follow-up, which mainly describe the growth process of odontogenic myxofibroma. We herein report a case of odontogenic myxofibroma that arose in the mandibular angle of a pediatric patient with a long-term follow-up of more than 14 years. The patient, when he was 10-years-old, first approached us with a complaint of painless swelling in the right mandibular angle that was palpable and showed bone-like hardness. Computed tomography revealed an ill-defined bone defect measuring 20 mm in diameter with bone protrusion in the margin. A biopsy was performed to differentiate the mass from osteosarcoma, giving a preliminary diagnosis of odontogenic myxofibroma or chondromyxofibroma. Therefore, tumorectomy was performed under general anesthesia. We curetted the bone surrounding the tumor and filled defect with iliac cancellous bone. The final diagnosis was odontogenic myxofibroma. At present, 14 years after the surgery, the patient had experienced an uneventful post-operative course without recurrence.

Keywords: Odontogenic myxofibroma; Pediatric tumor; Long-term follow-up

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1. Background

Odontogenic myxofibroma of the jawbone is a relatively rare disease with very few reports regarding its occurrence in children under 10 years of age.[1-3] In most cases, the lesion extends to the alveolar region and is accompanied by some dental symptoms such as tooth deviation and movement.[4] In addition, radiographic images may show significant bone destruction, resorption, and protrusion, which are features requiring differentiation from malignant diseases such as osteosarcoma.[5,6] Although post-operative recurrence has been reported to be common,[7,8] cases reporting long-term follow-up of the growth process remain scarce.

We herein report a pediatric case of odontogenic myxofibroma confined to the mandibular angle, who had experienced an uneventful postoperative course with no recurrence after more than 14 years of follow-up.

2. Case presentation

In June 2009, a 10-year-old Japanese boy presented to our department following a complaint of a painless swelling in the right mandibular angle. The pediatric patient had no remarkable personal or family medical history. Clinical findings included a palpable bone-like hard swelling in the right mandibular angle, but there were no abnormal findings in the oral cavity.

Panoramic X-ray and computed tomography (CT) scan revealed an ill-defined radiolucent image measuring 20 mm in diameter with an indistinct border, featuring a bone-like protrusion in the right mandibular angle (Figures 1 and 2). Magnetic resonance imaging revealed low signal intensity on T1-weighted imaging and high signal intensity on T2-weighted short inversion time inversion-recovery imaging with a partially irregular border with the surrounding bone (Figure 3). Because the possibility of a malignant tumor could not be ruled out merely based on the imaging findings, a biopsy was performed, and a histological evaluation of the biopsied sample rule out a diagnosis of osteosarcoma in our patient. The histopathological findings demonstrated fibrous proliferation with bland spindle cells, and the immunostaining showed negative expression of AE1/AE2 and desmin, and positive expression of SMA and bcl2. The diagnosis was deemed to be odontogenic myxofibroma or chondromyxofibroma.



Figure 1. A panoramic X-ray scan. An ill-defined bone defect with cortical bone resorption and the outward protrusion of the surrounding cortical bone-like spicules could be seen in the right mandibular angle (arrows).

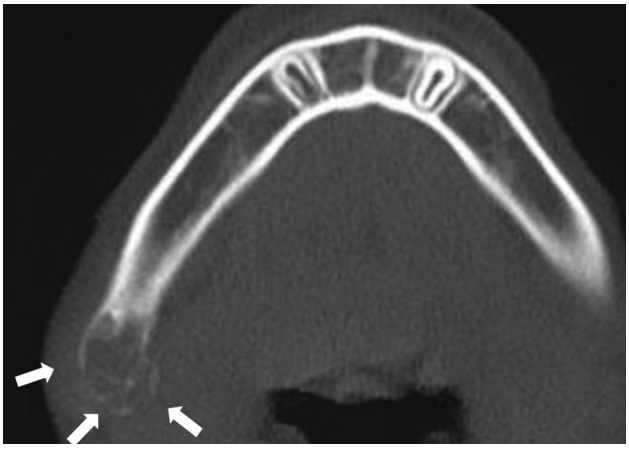


Figure 2. Computed tomography image obtained in the axial plane. A bone-like spicule was observed around the cortical bone defect with an unclear border (arrows).

In October 2009, the patient underwent tumorectomy and grafting of iliac cancellous bone under general anesthesia (Figure 4). A surgery was operated on the submandibular lesion, and the tumor was enucleated in one lump. Although the border between the tumor and bone was relatively clear, the bony surface exposed after tumorectomy was curetted in one layer with a rounded carbide bur. An absorbable plate was applied to give the mandibular angle morphology, and the gap between the plate and defect was filled with iliac cancellous bone. Macroscopically, the tumor was an elastic, soft lesion measuring 17 × 15 × 10 mm in size with a glossy surface and translucent white color (Figure 5).

Histopathologically, there was no obvious odontogenic epithelium, and spindle-shaped cells with little atypia were found to have proliferated in the mucous-like matrix. Immunostaining showed that AE1/AE2, desmin, and S-100 were negative, while SMA was positive. These findings support a final diagnosis of odontogenic myxofibroma (Figure 6).

Panoramic X-ray at 8 years postoperatively showed that the grafted bone had been integrated with the residual bone, without obvious difference in mandibular growth between the left and right sides (Figure 7). More than 14 years since the surgery had passed, the post-operative course for the patient had been uneventful without recurrence.



Figure 3. Magnetic resonance imaging. (A) T1-weighted image (axial plane). (B) T2-weighted short inversion time inversion-recovery image (axial plane). T1-weighted image showed a low signal intensity (arrows), and T2-weighted image showed a high signal intensity with a partially irregular border with the surrounding bone (arrows).

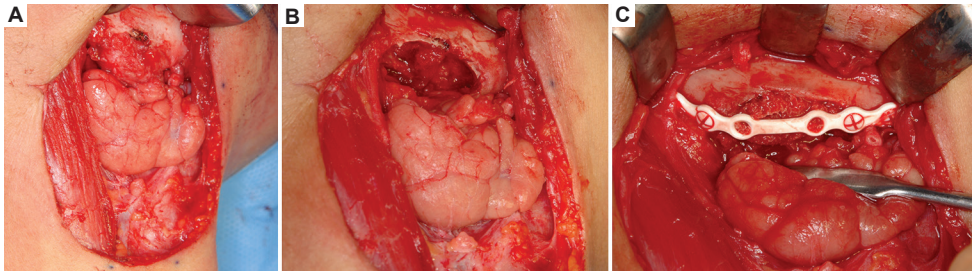


Figure 4. Intraoperative findings. (A) Surgical exposure of the lesion. (B) The lesion after tumorectomy. (C) The lesion after iliac cancellous bone grafting. An absorbable plate was applied to give the mandibular angle morphology, and the gap between the plate and defect was filled with iliac cancellous bone.

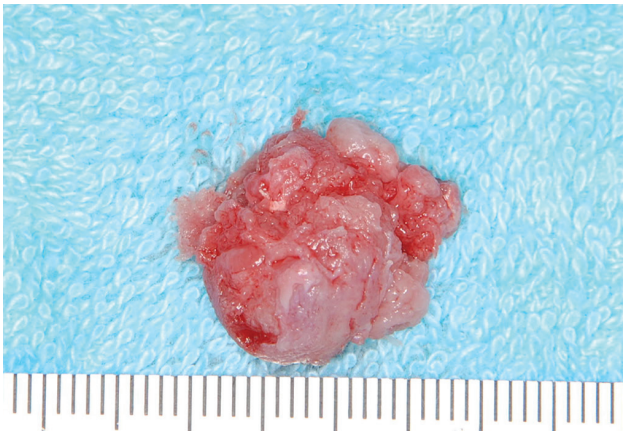


Figure 5. Enucleated specimen. The enucleated specimen, with a glossy surface and translucent white color, measures 17 × 15 × 10 mm in size.

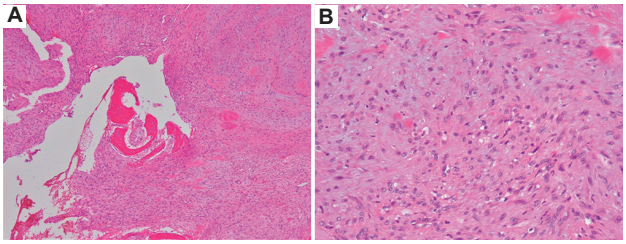


Figure 6. (A and B) Histopathological findings (hematoxylin-eosin stain). Both the image at different magnification showed. spindle-shaped cells with little atypia had proliferated in the mucous-like matrix, and the presence of odontogenic epithelium was unclear.

3. Discussion

Odontogenic myxofibromas are relatively rare tumors that occur in the jawbone intraosseously. In the 2017 World Health Organization classification of odontogenic tumors,[9] benign odontogenic tumors are classified into three categories: Benign epithelial odontogenic tumors, benign mixed epithelial-mesenchymal odontogenic tumors, and benign mesenchymal odontogenic tumors. Odontogenic myxofibroma is categorized as a benign mesenchymal odontogenic tumor, like odontogenic myxoma. Among odontogenic myxomas, those with a lot of fibrous formation



Figure 7. A panoramic X-ray scan at 8-year postoperatively. The grafted bone at the right mandibular angle had been integrated with the existing bone, and the mandibular angle morphology was found to be under recovery (arrows).

in the mucous-like matrix are specifically diagnosed as odontogenic myxofibromas. Typically, lesions that occur in the jawbone are defined as odontogenic, but such a definition does not extend to the presence or absence of an odontogenic epithelial mass while making a diagnosis.[1,4,10] Indeed, many cases have actually been reported in which no odontogenic epithelium is found, as in our present case (Tables 1 and 2).

The incidence of both odontogenic myxoma and odontogenic myxofibroma is estimated to be 3.3 – 15.7% among odontogenic tumors in Europe and the US.[5,7] The incidence in children under 10 years of age is 0.7 – 3.5%, which is lower than that in adults.[1-3] In Japan, the incidence of odontogenic myxoma and myxofribroma is 1.6 – 2.7%, which is lower than that in other countries.

Since statistics on odontogenic myxofibromas alone are not widely available, we performed a clinical statistical analysis of 46 cases of odontogenic myxofibromas of the mandible occurring in Japan from 1970 to 2023. The patient age ranged from 3 to 64 years old, and 38 cases (82.6%) were ≥11 years old (Table 1), while 8 (17.4%) were ≤10 years old (Table 2). There was a slight preponderance of female patients in the cases, with 17 male cases (44.7%) and 21 female cases

Table 1. Reports of odontogenic myxofibroma in the mandible angle in Japanese cases aged ≥11 years old

Case	Authors	Year	Age (years)	Gender	Radiographic findings				Localization	Treatment	Odontogenic epithelium	Follow-up time (months)
					Features	Maximum diameter of lesion (mm)	Alveolar bone invasiveness	Unerupted impacted tooth				
1	Momiyama *et al.*[11]	1970	13	F	Honeycomb	80	Yes	Yes	The right mandibular first premolar~second molar	Enucleation and curettage	No	Unknown
2	Masuda *et al.*[12]	1971	36	F	Multilocular; clear border	50	Yes	No	The left mandibular canine~third molar	Partial mandibulectomy	No	51
3	Watanabe *et al.*[13]	1971	25	M	Multilocular; dendriform	30	Yes	No	The left mandibular first premolar~first molar	Enucleation and curettage	No	5
4	Okayama *et al.*[14]	1973	27	F	Honeycomb; dendriform; unclear border	50	Yes	Yes	The right mandibular first molar~the right mandibular ramus	Enucleation and curettage	No	Unknown
5	Sakai *et al.*[15]	1976	28	M	Unknown	Unknown	Unknown	No	The right mandibular canine~the left second premolar	Unknown	Unknown	Unknown
6			35	F	Unknown	Unknown	Unknown	No	The right mandibular second molar	Unknown	Unknown	Unknown
7			38	F	Unknown	Unknown	Unknown	No	The right mandibular second premolar~third molar	Unknown	Unknown	Unknown
8			44	M	Unknown	Unknown	Unknown	No	The left mandibular first molar	Unknown	Unknown	Unknown
9			27	F	Unknown	Unknown	Unknown	No	The right mandibular first molar	Unknown	Unknown	Unknown
10			43	F	Unknown	Unknown	Unknown	No	The right mandibular second premolar~second molar	Unknown	Unknown	Unknown
11	Okui *et al.*[16]	1976	32	F	Multilocular; clear border	20	Yes	No	The right mandibular central incisor~canine	Enucleation and curettage	No	Unknown
12	Izumi *et al.*[17]	1977	49	M	Dendriform	15	Yes	No	Unknown	Enucleation and curettage	No	72
13	Kurachi *et al.*[18]	1977	29	F	Multilocular; clear border	35	Yes	No	The right mandibular lateral incisor~the left mandibular canine	Partial mandibulectomy	No	48
14	Nishijima *et al.*[19]	1978	26	F	Multilocular; Dendriform; clear border	40	Yes	Yes	Unknown	Enucleation and curettage	No	24
15	Nishijima *et al.*[20]	1978	34	F	Multilocular; clear border	40	No	No	The left mandibular central incisor~second molar	Hemi-mandibulectomy	No	Unknown

(Cont'd...)

Table 1. *(Continued)*

Case	Authors	Year	Age (years)	Gender	Radiographic findings				Localization	Treatment	Odontogenic epithelium	Follow-up time (months)
					Features	Maximum diameter of lesion (mm)	Alveolar bone invasiveness	Unerupted impacted tooth				
16	Osaki *et al.*[21]	1978	48	M	Multilocular; clear border	20	Yes	No	The right mandibular first premolar~the left mandibular first premolar	Enucleation and curettage	No	9
17	Furuta *et al.*[22]	1979	17	M	Ground glass appearance; slightly unclear border	Unknown	Yes	No	The right mandibular ramus	Enucleation and curettage	No	24
18	Abe *et al.*[23]	1979	44	F	Multilocular; clear border	43	Yes	No	The right mandibular canine~the left mandibular first premolar	Enucleation and curettage	Yes	6
19	Miyake *et al.*[24]	1981	35	F	Multilocular; clear border	30	Yes	No	The right mandibular first molar~the right mandibular ramus	Partial mandibulectomy	No	30
20	Tanaka *et al.*[25]	1983	64	M	Multilocular	90	Yes	No	The left mandibular first molar~third molar	Hemi-mandibulectomy PCBM grafts (plate reconstruction)	No	13
21	Futenma *et al.*[26]	1984	16	M	Clear border	65	Yes	Yes	The left mandibular first premolar~first molar	Enucleation and curettage	Yes	28
22	Osuga *et al.*[27]	1985	43	M	Multilocular; Dendriform; unclear border	77	Yes	No	The left mandibular canine~the left mandibular notch, condylar process	Segmental mandibulectomy PCBM grafts (plate reconstruction)	No	30
23	Nomura *et al.*[28]	1987	29	F	Multilocular; dendriform; slightly unclear border	50	Yes	No	The left mandibular second premolar~third molar	Enucleation and curettage	No	15
24			43	F	Multilocular; partially irregular border	25	Yes	No	The right mandibular canine~second molar	Enucleation and curettage	No	10
25	Ohshima *et al.*[29]	1987	25	M	Sun-ray appearance; partially clear border	20	Yes	No	The left mandibular first premolar~second molar	Enucleation and curettage	Yes	8
26	Yasukawa *et al.*[30]	1988	15	F	Multilocular; dendriform; unclear border	70	Yes	Yes	The right mandibular first premolar~first molar	Segmental mandibulectomy PCBM grafts (plate reconstruction)	No	About 48
27			43	M	Bilocular; clear border	20	Yes	No	The left mandibular first premolar~first molar	Enucleation and curettage PCBM grafts	No	17

(Cont'd...)

Table 1. *(Continued)*

Case	Authors	Year	Age (years)	Gender	Radiographic findings				Localization	Treatment	Odontogenic epithelium	Follow-up time (months)
					Features	Maximum diameter of lesion (mm)	Alveolar bone invasiveness	Unerupted impacted tooth				
28			24	F	Dendriform; clear border	60	Yes	Yes	The right mandibular second premolar~the right mandibular ramus	Segmental mandibulectomy PCBM grafts (plate reconstruction)	No	27
29	Ishihara *et al.*[31]	1989	26	M	Unilocular; clear border	30	Yes	No	The right mandibular first premolar~first molar	Enucleation and curettage	No	24
30	Okada *et al.*[32]	1990	26	M	Unilocular; clear border	30	Yes	Yes	Unknown	Enucleation and curettage	Yes	18
31	Nakagawa *et al.*[33]	1998	37	M	Multilocular; clear border	80	Yes	Yes	The right mandibular second premolar~the right mandibular ramus	Segmental mandibulectomy	Yes	8
32	Suzuki *et al.*[34]	1999	24	F	Soap bubble appearance	44	Yes	No	The right mandibular first premolar~the left mandibular lateral incisor	Enucleation and curettage	Yes	12
33	Shimizu *et al.*[35]	2004	61	F	Multilocular; clear border	30	Yes	No	The left mandibular second premolar~third molar	Segmental mandibulectomy PCBM grafts (plate reconstruction)	No	16
34	Yagishita *et al.*[36]	2007	41	M	Multilocular; clear border	40	Yes	No	The left mandibular median~the left mandibular coronoid process	Enucleation and curettage	No	17
35	Kadono *et al.*[37]	2009	41	M	Multilocular; dendriform; clear border	60	Yes	Yes	Unknown	Enucleation and curettage	Yes	18
36	Suwa *et al.*[38]	2013	20	F	Multilocular; clear border	10	Yes	No	The right mandibular lateral incisor~second premolar	Enucleation and fenestration	No	24
37	Ohta *et al.*[39]	2015	36	F	Clear border	Unknown	Yes	No	The right mandibular second premolar~the right mandibular ramus	Segmental mandibulectomy; mandibular reconstruction using PCBM grafts with titanium mesh tray	No	72
38	Sato *et al.*[40]	2018	22	M	Unilocular; clear border	30	Yes	No	The left mandibular third molar~the left condyle	Zygomatic osteotomy; segmental mandibulectomy; plate reconstruction	No	84

Abbreviations: M: Male; F: Female; PCBM: Particulate cancellous bone and marrow.

Table 2. Reports of odontogenic myxofibroma in the mandible angle in Japanese cases aged ≤10 years old

Case	Authors	Year	Age (y)	Gender	Radiographic findings				Localization	Treatment	Odontogenic epithelium	Follow-up time (months)
					Features	Maximum diameter of lesion (mm)	Alveolar bone invasiveness	Unerupted impacted tooth				
1	Ito *et al.*[41]	1973	10	M	Unilocular clear border	80	Yes	Yes	The left mandibular second molar~the left mandibular ramus	Enucleation and curettage PCBM grafts	No	8
2	Inoue *et al.*[42]	1979	7	F	Unilocular	43 and 20 Two lesions	Yes	Yes	The left mandibular second primary molar~the left mandibular ramus	Enucleation and curettage	Yes	24
3	Osuga *et al.*[27]	1985	8	F	Dendriform; honeycomb; clear border	27	Yes	No	The right mandibular second primary molar	Enucleation and curettage	No	20
4	Sato *et al.*[43]	2017	8	M	Unilocular; clear border	20	Yes	Yes	The right mandibular second molar	Enucleation and curettage	Yes	42
5	Kanda *et al.*[44]	2018	6	M	Unilocular; clear border	25	Yes	Yes	The left mandibular second primary molar~first molar	Enucleation and fenestration	Yes	24
6	Oda *et al.*[45]	2020	8	M	Unilocular; clear border	Unknown	Yes	Yes	The right mandibular first molar~the right mandibular ramus	First biopsy: fenestration; second biopsy: enucleation and curettage	No	44
7	Kanoh *et al.*[46]	2021	3	F	Unilocular; clear border	16	No	No	The right mandibular angle	Enucleation and curettage	No	84
8	The current case	2023	10	M	Unilocular; unclear border	17	No	No	The right mandibular angle	Enucleation and curettage PCBM grafts (plate reconstruction)	No	168

Abbreviations: M: Male; F: Female; PCBM: Particulate cancellous bone and marrow.

(55.3%), which is consistent with previous reports.[4,8] However, among children ≤10 years of age, 5 (62.5%) were boys and 3 (37.5%) were girls, highlighting a slight preponderance of male patients. In general, the lesions invade into the alveolar bone and are often accompanied by root resorption and deviation of adjacent teeth. Only three cases (6.5%), including our own, were reported to be localized in the mandibular angle and not associated with teeth.

Differential diagnoses include ameloblastoma, myxoid nerve sheath tumors, chondromyxoid fibroma, low-grade myxofibrosarcoma,[9] and osteosarcoma. Among them, differentiation from malignancy is of significant clinical relevance. The radiographic and CT findings in myxofibromas are often uni- or multi-locular radiolucent images showing unclear borders, and the cortical bone around the lesion is usually bulging and thinning.[47] In the present case, the radiographic and CT images showed a unilocular radiolucent image with unclear borders accompanied by cortical bone resorption and protrusion. This finding was similar to the characteristic of osteosarcoma of the jaw, such as osteoid protrusion (spicula) and reactive trabecular bone formation (sun-ray image), which made differentiation difficult.[6] In some cases of osteosarcoma of the jawbone, radiographic findings have shown a radiolucent image with a clear border, such as a cyst, which reportedly hampered differentiation from benign diseases.[48,49] Therefore, making a careful diagnosis is critical.

Radical resection or conservative surgery is the most common method of treatment, and Meleti *et al.*[8] stated that the rates were 26.09% and 73.91%, respectively. In the present analysis of 46 cases in Japan, radical resection was performed in 12 cases (26.1%) and conservative surgery in 28 cases (60.9%), with the approach unknown in 6 cases (13.0%), a trend similar to that reported in Europe and the US.[8] There are differences of opinion concerning the selection of surgical procedure, with some reports supporting the choice of radical resection in cases of large lesions or recurrence, considering the local invasiveness of the tumor.[1,50,51] However, extensive jaw resection in children may induce aesthetic issues and psychological suffering due to hindered growth of the maxillofacial region and a deformed facial appearance. Therefore, it is suggested that benign mandibular tumors in young patients first be treated with enucleation and curettage, even if the tumor is large enough to be considered for jaw resection. In fact, in the eight pediatric cases ≤10 years of age assessed in this analysis, enucleation and curettage or enucleation and fenestration were selected, and conservative surgery was performed in 100% of these cases. In contrast, in 38 adult cases, conservative treatment was applied in only 20 cases (52.6%), suggesting that conservative surgery was more commonly performed in children than in adults. In this case study, we performed enucleation and curettage, considering the site of origin, age, growth, and post-operative appearance. The patient had been well without recurrence for 14 years since the surgery. Until now, there have been no functional or aesthetic issues reported by the patient.

Odontogenic myxofibromas may show invasive growth into bone, and there are reports of recurrence in 9.5% of patients over 3 years of follow-up.[8,52] In children, 20% of myxomas and myxofibromas in patients under 16 years of age recurred within 1 year after surgery.[7] There were no reports of recurrence among the 46 Japanese cases we analyzed. However, the mean post-operative observation period of the 45 patients, excluding our own case, was only 2 years and 3 months (ranging from 5 months to 7 years), and data on post-operative observation period were not described for 10 cases. In addition, the follow-up period of the six pediatric cases ranged from 8 months to 7 years, and none of the cases had a long-term follow-up period as long as 14 years like ours. Thus, the actual recurrence rate of odontogenic myxofibromas in the long term is unknown. A large-scale study of the long-term prognosis should be conducted to investigate the appropriateness of conservative surgery. In addition, given that studying post-operative growth is taken into account in pediatric cases, we consider that long-term follow-up is necessary to evaluate the functional and esthetic effect of the procedure.

4. Conclusion

We report a case of odontogenic myxofibroma that arose in the mandibular angle of a pediatric patient with a long-term follow-up of more than 14 years. The occurrence of odontogenic myxofibromas in the jawbone of children has been rarely reported.[53] This case report and literature statistics will provide helpful information for the diagnosis and determining treatment strategies for odontogenic myxofibroma in children with growth process.

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Conflict of interest

The authors declare no conflicts of interest.

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Ethics approval and consent to participate

An explicit permission or consent of the case subject described in this case report had been obtained before their participation.

Consent for publication

Informed consent has been obtained from the patient for publication of this case report.

Availability of data

The data can be requested from the corresponding author following reasonable request.

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