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Case Report

Evaluating for systemic artery aneurysms using noncontrast magnetic resonance angiography in patients with Kawasaki disease: A report of two cases ☆,☆☆

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ARTICLE INFO

Article history:

Received 28 October 2020

Revised 23 December 2020

Accepted 25 December 2020

Keywords:

Magnetic resonance imaging

Kawasaki disease

Systemic artery aneurysms

Noncontrast magnetic resonance angiography

ABSTRACT

Kawasaki disease (KD) involves coronary aneurysms and can infrequently cause systemic artery aneurysms (SAAs). Therefore, patients with KD should be evaluated for both coronary and systemic arterial aneurysms. This report describes 2 cases of SAA evaluated using the diastolic phase image of electrocardiogram-gated three-dimensional fast spin echo during noncontrast magnetic resonance angiography. The first case was a 1-year-old male who diagnosed with KD at 2 months of age. Multiple right axillary artery aneurysms measuring 6.0 mm and 2.5 mm and left axillary artery aneurysms measuring 12.0 mm, 4.0 mm, and 3.0 mm were observed by scanning for 94 seconds. The second case was a 13-year-old male who diagnosed with KD at 4 months of age, with a 7.0-mm right axillary artery aneurysm observed by scanning for 101 seconds. Electrocardiogram-gated three-dimensional fast spin echo in the diastolic phase can help evaluate SAA in patients with KD and does not require a prolonged scanning time or contrast medium.

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☆ Funding: This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

☆☆ Competing interests: Masami Yoneyama is an employee of Philips Japan. Other authors have no conflicts of interests.

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<https://doi.org/10.1016/j.radcr.2020.12.062>

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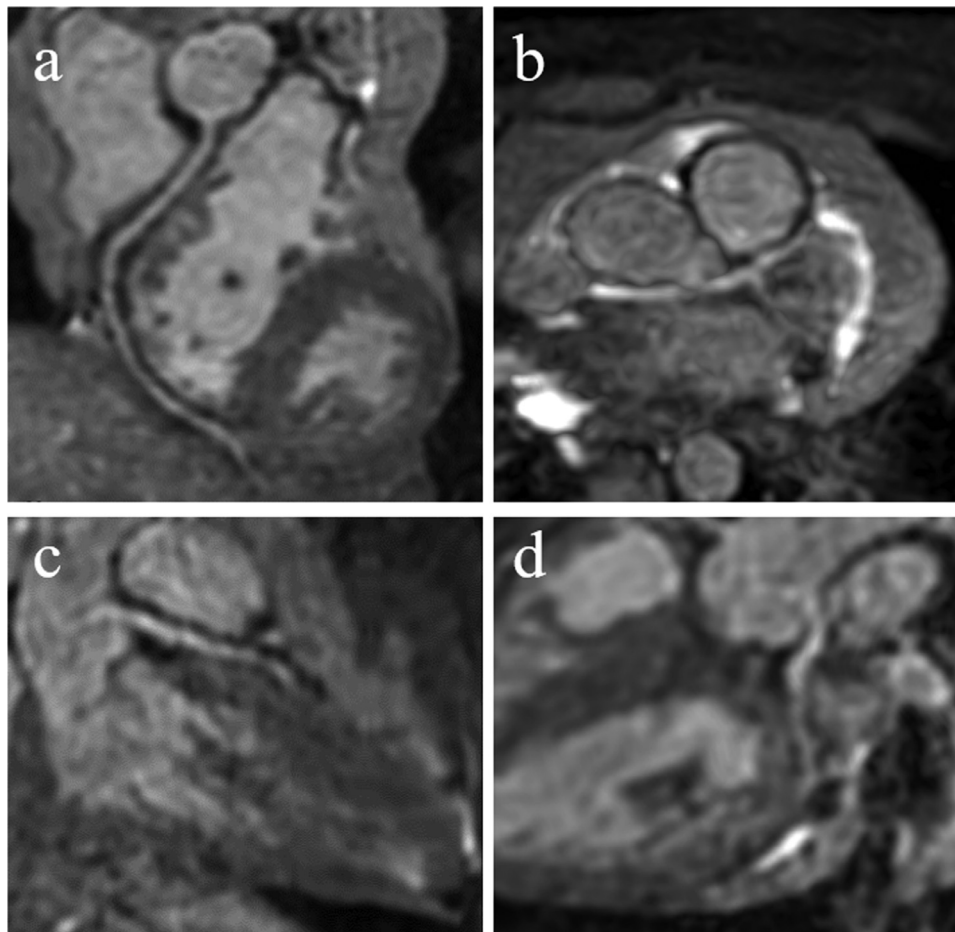


Fig. 1 – Coronary arteries in a 1-year-old male with Kawasaki disease. Maximum intensity projection images using the ECG and the navigator gated by using 3-dimensional steady state free precession. In coronary MRA, all coronary aneurysms tended to regress. (a) The right coronary artery for 2.2 mm (Z score = +2.1) (b) The left main trunk for 2.0 mm (Z score = +0.6) (c) The left anterior descending coronary artery for 1.6 mm (Z score = +0.4) (d) The left circumflex coronary artery for 1.6 mm (Z score = +0.6).

Introduction

Kawasaki disease (KD) involves acute febrile systemic vasculitis. The etiology of KD is unknown. If the coronary arteries are involved, ongoing treatment or surgery may be required. Without treatment, coronary artery aneurysms occur in up to 25% and lead to death in about 1% cases [1]. KD particularly affects the coronary arteries; however, in approximately 2% of untreated patients, KD also causes systemic artery aneurysms (SAA) [1]. Severe SAA is associated with ruptured iliac aneurysms [2], axillary artery occlusion [3], and artificial blood vessel replacement for thoracic aneurysms [4]. Therefore, patients with KD should be evaluated for both coronary and systemic arterial aneurysms.

Various modalities such as ultrasonography (US) [5], angiography (AG) [1], computed tomography angiography (CTA) [4], and contrast-enhanced magnetic resonance angiography (CE-MRA) [2] are used to evaluate SAAs in patients with KD. However, the optimal method for evaluating SAA has not been established. Therefore, there is a need to optimize minimally invasive methods for evaluating SAA in patients with KD.

Recently, noncontrast magnetic resonance angiography (NC-MRA) has been used clinically. One method for NC-MRA involves electrocardiogram (ECG)-gated 3-dimensional fast spin echo (3D-FSE). A previous report indicated that evaluation of the aorta was possible using the diastolic phase image of ECG-gated 3D FSE [6]. However, there are no reports of using this method to evaluate SAA during the diastolic phase with ECG-gated 3D FSE. This report describes the successful use of ECG-gated 3D FSE in the diastolic phase to evaluate SAA in 2 patients with KD.

Case 1

The first case was a 1-year-old male who at 2 months of age, presented with high fever, conjunctival congestion, changes in the lips and oral cavity, polymorphous rash, and cervical lymphadenopathy. He was diagnosed with KD and received a dose of intravenous immunoglobulin. The right coronary artery (RCA) aneurysm measured 4.3 mm, the left main trunk (LMT) aneurysm measured 3.4 mm, the left anterior descending coronary artery (LAD) aneurysm measured 2.5 mm, and

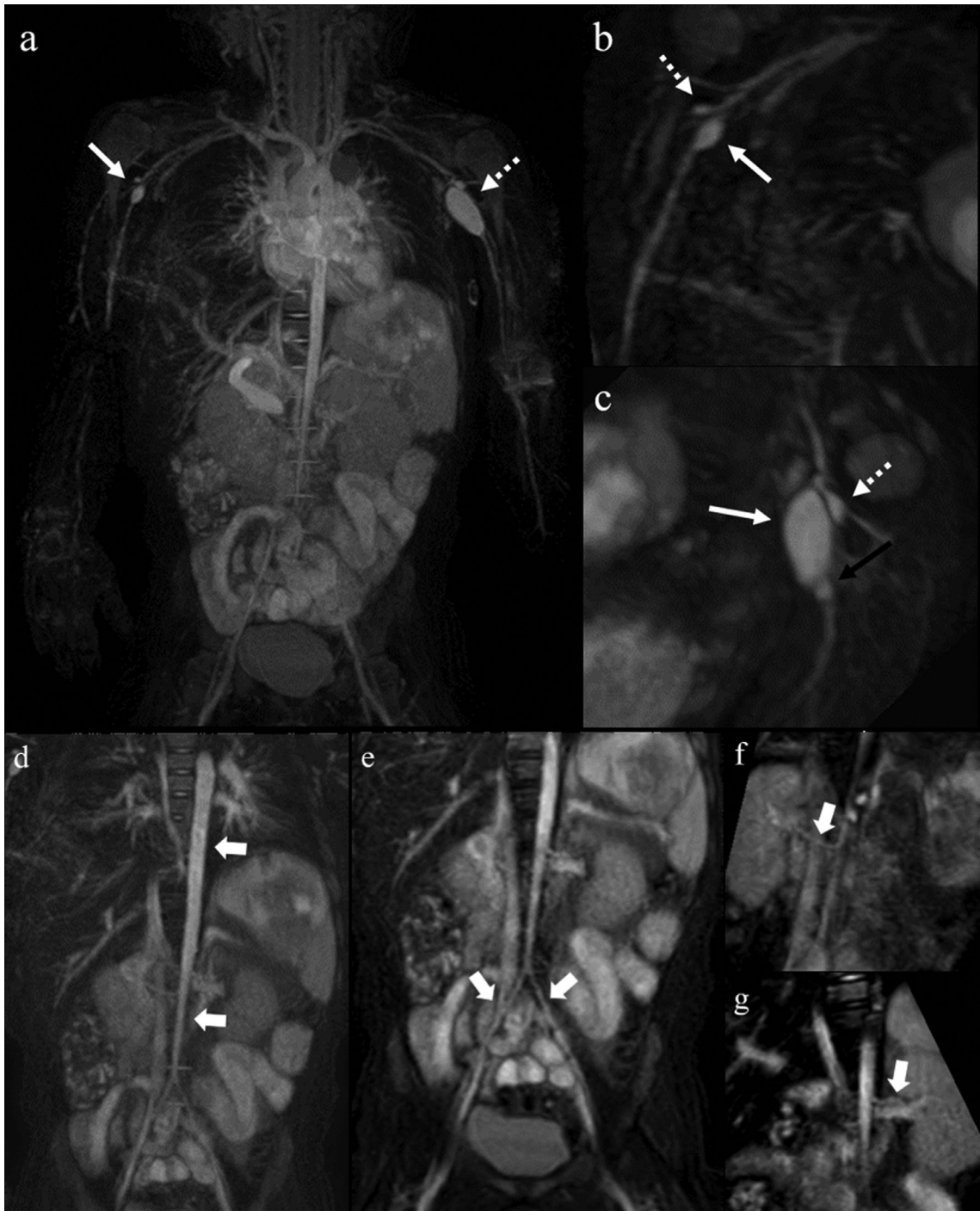


Fig. 2 – Bilateral axillary artery aneurysms in a 1-year-old male with Kawasaki disease maximum intensity projection images using the Electrocardiogram-gated 3-dimensional fast spin echo in the diastolic phase revealed multiple right axillary artery aneurysms and multiple left axillary artery aneurysms with a scan time of 94 seconds (a). The right axillary artery aneurysms (b) were 6.0 mm (solid arrows) and 2.5 mm (dashed arrows) in size. The left axillary artery aneurysms (c) were 12.0 mm (solid arrows), 4.0 mm (dashed arrows), and 3.0 mm (black arrows) in size. It was possible to evaluate for aneurysms in whole-body blood vessels such as descending aorta (d), iliac arteries (e), right renal artery (f), and left renal artery (g), in addition to the axillary arteries.

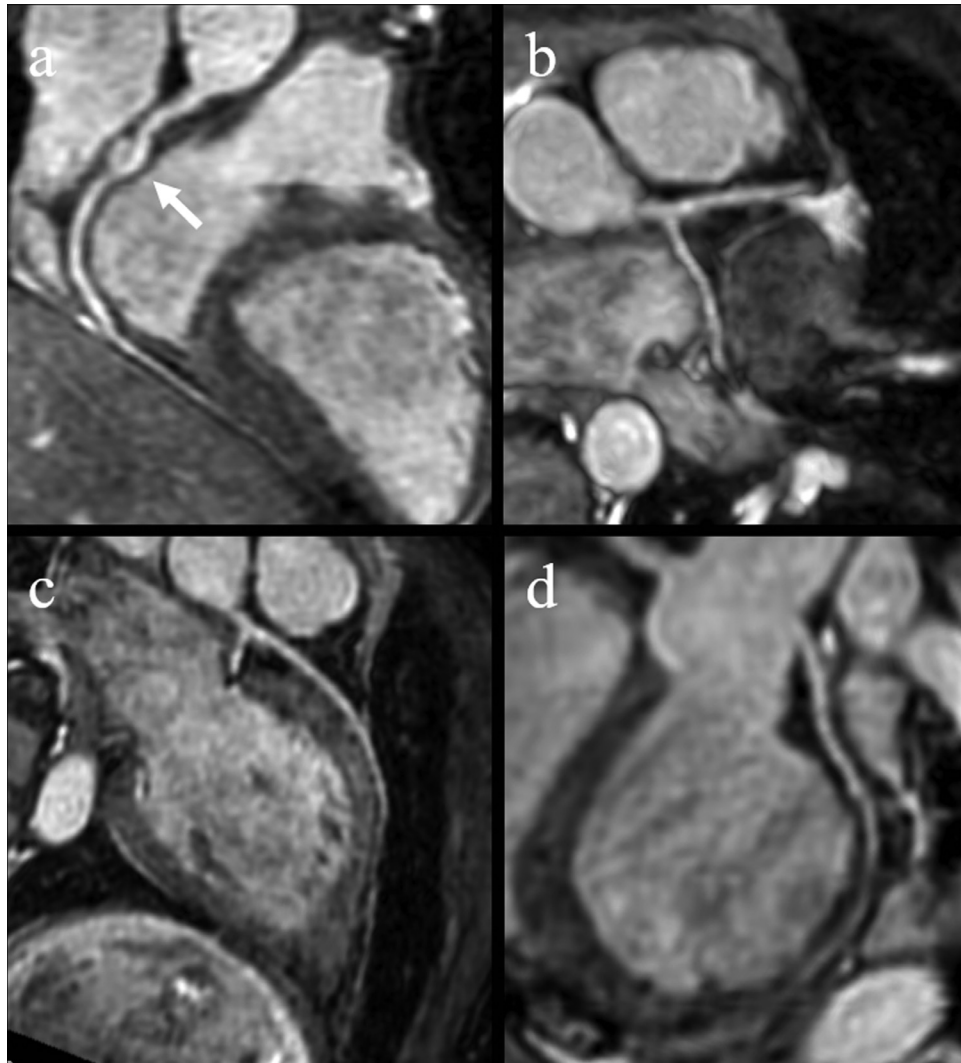


Fig. 3 – Coronary arteries in a 13-year-old male with Kawasaki disease. Maximum intensity projection images using the ECG and the navigator gated by using three-dimensional steady state free precession. In coronary MRA, RCA aneurysm for 6.7mm (Z score = +6.9) was observed. (a) The right coronary artery for 6.7 mm (Z score = +6.9) (b) The left main trunk for 3.2 mm (Z score = +0.5) (c) The left anterior descending coronary artery for 2.7 mm (Z score = +0.6) (d) The left circumflex coronary artery for 2.3 mm (Z score = +0.3).

the left circumflex coronary artery (LCX) aneurysm measured 3.2 mm were identified in the US. The Z score evaluates the internal diameter of the coronary arteries in patients with KD and represents the degree of coronary aneurysm. A coronary artery Z score between +2.0 and less than +2.5 (ie, 2 to less than 2.5 standard deviations above the average normalized for body surface area) is considered dilated. A coronary artery with a Z score between +2.5 and less than +5.0 is considered a small aneurysm. A Z score between +5.0 and less than +10.0 is considered a large aneurysm. A Z score of +10.0 or greater is considered a giant aneurysm [7]. The Z scores were +8.0 for RCA, +5.8 for LMT, +4.2 for LAD, and +4.9 for LCX. After 11 months, NC-MRA was performed by sedation with thiopental sodium (Ravonal, Mitsubishi Tanabe Pharma Co., Osaka, Japan) 40 mg to evaluate the coronary arteries and SAA.

We used the 1.5-T MRI clinical imager (Ingenua, Philips Healthcare, Best, the Netherlands) using Flex S coil and poste-

rior coil. The imaging parameters were as follows: repetition time, 1090.0 ms (2 beats); echo time, 75 ms (shortest); field of view, 380 mm; matrix, 320 × 256; slice thickness, 3.6 mm; reconstruction slice thickness, 1.8 mm; number of slices, 40; flip angle, 90°; refocusing flip angle, 180°; echo train length, 45; inversion time, 165 ms; sensitivity encoding factor, 1.5; half scan factor, Y 0.8, Z 1.0; trigger delay, 791.4 ms; and scan time, 94 seconds. ECG-gated 3D FSE in the diastolic phase image was acquired with coronal sections. Coronary MRA was performed with ECG and the navigator-gated acquisition time was approximately 7 minutes.

In coronary MRA, the RCA aneurysm and LMT aneurysm regressed to 2.2 mm and 2.0 mm, respectively. The Z scores were +2.2 for RCA and +2.0 for LMT (Fig. 1). However, multiple axillary aneurysms were identified—6.0-mm and 2.5-mm right axillary artery aneurysms and 12.0-mm, 4.0-mm, and 3.0-mm left side axillary artery aneurysms (Fig. 2). There were



Fig. 4 – Right axillary artery aneurysms in a 13-year-old male with Kawasaki disease maximum intensity projection images using the Electrocardiogram -gated 3-dimensional fast spin echo in the diastolic phase was able to observed right axillary artery aneurysm with a scan time of 101 seconds. (a) (solid arrows). Note the 7.0-mm right axillary artery aneurysms (b) (solid arrows). It was possible to evaluate for aneurysms in whole-body blood vessels such as the descending aorta and iliac arteries (c), axillary artery (d), right double renal artery (e), and left double renal artery (f).

no aneurysms in the other trunk arteries. It was possible to evaluate the descending aorta, iliac artery, right renal artery, and left renal artery using the ECG-gated 3D FSE in the diastolic phase (Fig. 2).

Case 2

The second case was a 13-year-old male. At 4 months of age, he presented with a fever, conjunctival congestion, changes in the lips and oral cavity, polymorphous rash, and cervical lymphadenopathy. He was subsequently diagnosed with KD and received a dose of intravenous immunoglobulin. RCA aneurysm measuring 8.0 mm, LMT–LAD aneurysm measuring 4.0 mm, and axillary artery aneurysm measuring 5.0 mm were identified by US. The Z scores were +13.3 for RCA and +7.1 for LMT–LAD. After 13 years, NC-MRA was performed by sedation with thiopental sodium 240 mg to evaluate the coronary arteries and SAA.

We used a torso coil. The imaging parameters were as follows: repetition time, 1500 ms (2 beats); echo time, 70 ms (shortest); field of view, 500 mm; matrix, 320 × 256; slice thickness, 3.6 mm; reconstruction slice thickness, 1.8 mm; number of slices, 50; flip angle, 90°; refocusing flip angle, 180°; echo train length, 45; inversion time, 165 ms; sensitivity encoding-factor, 2.0; half scan factor, Y 0.8, Z 1.0; trigger delay, 1176 ms; scan time, 101 seconds. ECG-gated 3D FSE in the diastolic phase image was acquired with coronal sections. Coronary MRA was performed with ECG, and the navigator-gated acquisition time was approximately 8 minutes.

In coronary MRA, the LMT–LAD aneurysm regressed to 3.2 mm (Fig. 3) but the RCA aneurysm regressed to 6.7 mm. The Z scores were +6.9 for RCA and +0.5 for LMT–LAD (Fig. 3). A residual right axillary artery aneurysm measuring 7.0 mm was observed (Fig. 4); however, no changes—such as new stenotic lesions—were observed. It was possible to evaluate the descending aorta, iliac artery, subclavian artery, axillary artery, right renal artery, and left renal artery using the ECG-gated 3D FSE in the diastolic phase. In this case, bilateral renal arteries were double renal arteries and were well detected (Fig. 4).

Discussion

ECG-gated 3D FSE in the diastolic phase of NC-MRA is useful for evaluating SAAs in patients with KD. This method can capture both arteries and veins with high signals by acquiring images during the slow blood flow period of the diastole phase. Because there is no inflow effect, it allows for coronal 3D acquisition. Compared with the image method in which transverse images of blood vessels are acquired using the inflow effect, it is possible to perform short-time imaging with a few slices. This report imaging was completed within 2 min. Given that this was possible in such a short time, ECG-gated 3D FSE in the diastole phase was acquired as an additional imaging modality to noncontrast coronary MRA. The coronary arteries and SAA can be evaluated in one examination using NC-MRA. This is ideal for patients with KD.

Also, ECG-gated 3D FSE in the diastolic phase is less affected by magnetic field inhomogeneity because the FSE method uses multiple spin refocusing pulses and the short-tau inversion-recovery method using the inversion pulse is used for fat suppression. Therefore, it was possible to reduce the magnetic susceptibility of the air in the lung, acquire a wide area with fat suppression by homogeneous, and view the systemic arteries in two patients with KD.

It is assumed that SAA requires long-term follow-up after evaluation in the acute phase, similar to coronary aneurysms [8]. As children generally live longer than older individuals, their potential risk for developing cancer due to radiation exposure from diagnostic imaging is higher. There are a few reports of SAAs in patients with KD; however, the optimal screening method remains unknown. There are many modalities for evaluating SAA, such as US, AG, CTA, and CE-MRA [1,2,4,5]. US is not suitable for wide-range observation, and the images can vary widely relative to the observer's technique. AG and CTA are the most common modalities, but are invasive and involve radiation exposure and contrast medium. CE-MRA is also invasive and requires contrast medium; however, a recent report indicated that gadolinium agent accumulates in the brain [9]. ECG-gated 3D FSE in the diastole phase can accurately and noninvasively evaluate SAA. We reported a case of a 1-year-old with successful detection of SAA. This finding suggests that younger patients can also be evaluated.

Our report has some limitations. First, we were unable to evaluate SAA stenosis in patients with KD. SAA can lead to stenotic lesions in the late period, and evaluation of stenotic lesions is important. Second, these case findings have not been compared with CT or AG findings. Lastly, we did not evaluate the diagnostic accuracy of this method.

In conclusion, it is possible to evaluate SAA in patients with KD using ECG-gated 3D FSE in the diastole phase of NC-MRA.

Ethics approval

This study was approved by our institutional review board.

Consent to participate

Written informed consent was obtained from the parents.

Consent for publication

Written informed consent was obtained from the parents.

Availability of data and material

Not applicable.

Code availability

Not applicable.

Authors' contributions

All authors contributed to the study conception and design. Material preparation and data collection were performed by Haruki Nonaka. The first draft of the manuscript was written by Haruki Nonaka and Takanori Masuda and Takeshi Nakaura were commented on previous versions of the manuscript. All authors read and approved the final manuscript.

Supplementary materials

Supplementary material associated with this article can be found, in the online version, at doi:[10.1016/j.radcr.2020.12.062](https://doi.org/10.1016/j.radcr.2020.12.062).

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