

Interventional magnetic resonance imaging as a diagnostic and therapeutic method in treating acute pediatric atlantoaxial rotatory subluxation

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Abstract. Atlantoaxial rotatory subluxation or fixation (AARF) is a rare condition, usually occurring in pediatric patients. It mimics benign torticollis but may result in permanent disability or death. The condition requires prompt diagnosis by thorough examination to avoid any treatment delays. Spiral computed tomography (CT) with three-dimensional reconstruction CT is recommended for identifying incongruence between C1 and C2 vertebrae, and magnetic resonance imaging (MRI) may be performed to exclude ligamentous injuries. In addition to static imaging, dynamic CT involves the reduction between C1 and C2 being confirmed using CT with the head turned maximally to the left and right. The present report (level of evidence, III) provides a method for treating AARF that has similar advantages as dynamic CT but avoids ionizing radiation by replacing CT with interventional MRI. The new method comprised simultaneous axial traction and manual closed reduction, performed under general anesthesia, and the use of interventional MRI to ensure that reduction was achieved and held. The head is turned maximally to the right and left during the manual reduction. A rigid cervical collar was used following reduction. Dynamic CT was not required but prior diagnostic static CT was performed in preparation. No further CT was required. There appears to be no previous studies on interventional MRI in AARF care. Being superior in its diagnostic soft-tissue visualization performance and lacking ionizing radiation, interventional MRI is a potential option

for investigating and treating acute AARF in non-syndromic patients with no trauma history.

Introduction

Atlantoaxial rotatory fixation (AARF) is a condition in which the first two cervical vertebrae (C1 and C2) are fixed in rotational malalignment. The underlying cause of AARF remains to be fully elucidated (1). Mechanically, there is temporary atlantoaxial instability resulting in subluxation between C1 and C2, and failure to reduce spontaneously may lead to AARF (2). The condition is not unique to children, but it most frequently affects the growing skeleton. Subluxation may be associated with several underlying conditions, including acute acquired torticollis, clavicle fracture, upper-respiratory-tract infection, surgery in the head and neck area or minor trauma, although no identifiable predisposing factors are mandatory for the diagnosis. Long-term morbidity in cases of AARF is likely if the condition remains untreated, with instability in the atlantoaxial joint (3), and subsequent remodeling of the vertebrae to a non-anatomic form renders late spontaneous reduction impossible, highlighting the requirement for early surgical intervention (4). Missed AARF may result in paralysis, nerve damage and fatality (5).

Clinicians should always be aware of AARF when treating a child presenting with neck pain and an abnormal position of the head. Typically, the child is unable to return the head to a neutral position and turn it past the midline, but their neurological status is almost always normal. In cases where symptoms have lasted for more than a few days without spontaneous relief, AARF requires to be ruled out. However, the diagnosis of AARF is frequently delayed. Plain radiographs are difficult to interpret, albeit they are usually used in primary imaging for practical reasons (6-8). Computer tomography (CT) is valuable for measuring the rotatory angle and the distance between the atlas (C1) and dens (C2) (atlas-dens index), which reflects anterior displacement of the atlas (9). Children with benign torticollis or slight Fielding type 1 AARF present with a normal C1-C2 association in CT scans (6). Three-dimensional

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(3D) spiral CT is considered to be the best diagnostic tool to evaluate AARF (10-12). Dynamic CT covering the skull base and upper cervical spine with the head rotated to the maximal left and maximal right positions is recommended as a method of choice for the diagnosis of AARF, despite its poor reliability and reproducibility (7,13). Subluxation is classified into four groups, according to the CT findings (14): Type 1, unilateral anterior rotation of the atlas pivoting around the dens; type 2, unilateral anterior rotation of the atlas with an atlas-dens index of 3-5 mm; type 3, bilateral anterior subluxation with an atlas-dens index of >5 mm; type 4, posterior displacement of the atlas relative to the axis. Magnetic resonance imaging (MRI) of the brain and cervical spine is recommended for acquired torticollis if the CT findings are negative (9). MRI is superior in recognizing the potential associated ligamentous injuries in soft tissues (1).

It is known that early reduction of subluxation results in good outcomes and a low rate of recurrent subluxation, whereas subjects with a longer history of subluxation may require skeletal traction and halo/rigid collar immobilization or surgical reduction and fusion (15). Specifically, a Fielding type 2 (or higher) condition requires skeletal traction (16). Therefore, any neck pain and head malrotation persisting for more than a few days usually merit close investigation, i.e. static and/or dynamic CT scans. However, MRI has certain advantages over CT regarding the diagnostic performance and due to the lack of associated ionizing radiation (1). The present study reports on the pioneering practice of utilizing 1.5 Tesla (T) interventional MRI in the treatment of a pediatric patient with suspected acute AARF. In addition, the current literature regarding the imaging modalities used in cases of AARF for diagnostic purposes and during reduction was reviewed.

Materials and methods

Treatment strategy. In the present study, a radiation-free method for the treatment of AARF in a pediatric patient is described and it is based on interventional MRI instead of CT. The technique is explained in detail and its feasibility in the treatment of a young child was closely evaluated. The present study was based on the urgent *ad hoc* clinical treatment of the patient. It was considered, *a priori*, that this experimental study treatment method that utilizes interventional MRI instead of dynamic CT in confirming the reduction, would be safer with regard to radiation exposure and superior in the visualization of soft-tissue pathology than the traditional method using CT (1). Hence, ethics board approval was considered to not be required, also in the light of the poor condition of the patient and risks associated with delayed treatment, implying the urgency of intervention. However, besides interventional MRI, the treating team was still prepared to perform traditional care with using dynamic CT if required.

Literature review. A literature search was performed in the MEDLINE biomedical literature database, with an advanced search in PubMed using the following search terms: 'atlantoaxial rotation(al) fixation', 'atlantoaxial rotation(al) subluxation', 'nasopharyngeal torticollis' and 'Grisel syndrome'. The same terms were used for a search in the Google Scholar database, while the non-scientific citations retrieved were

ignored. Primarily, original studies and case reports in English and those with at least an English abstract were reviewed to study imaging practices and treatment techniques. Articles in languages other than English were excluded. Furthermore, beyond the systematic search described above, all reference lists of the published articles retrieved in these searches were reviewed to identify further relevant studies. The focus of the literature review was the role of static MRI in the early stage of diagnosis and the possible role in interventional investigation. Studies on patients with a growing skeleton and/or those of <16 years of age were considered eligible. Studies on fracture cases, syndromic patients and chronic AARF were excluded. The major focus of the present literature review was to identify imaging modalities used for diagnostic purposes in the acute stage of AARF and also to identify any published experience of using MRI as an interventional method during AARF reduction in children. The search included studies published between January 1st, 1998 and March 31st, 2018. Finally, a total of 28 original studies and case reports were included (Table I).

Results

AARF treatment approach using interventional MRI. As a major feature of the present study, a radiation-free orthopedic technique was developed. The treatment comprised of simultaneous manual axial traction and manual closed reduction of AARF, with the patient lying in the supine position under general anesthesia, with interventional MRI during the same period of anesthesia to make sure that reduction is achieved and held. For the present study, interventional MRI is classified as a therapeutic orthopedic procedure performed under special circumstance that allows unrestricted use of interventional MRI with viewing monitors close to the operating table and equipment for general anesthesia. The reduction maneuver is based on turning the head first maximally right and then left during traction, in a manner similar to that performed in the procedure called dynamic CT (13). Thereafter, a rigid cervical collar was used for several weeks to stabilize the vertebrae in the correct position.

Case presentation. A female patient aged 4 years with neck pain and the head tilted to the left for 1 week presented at the pediatric orthopedic unit Oulu University Hospital (Oulu, Finland; March 2018). The patient had not suffered any injury or accident and there was no sign of upper or lower-respiratory-tract infection. Movement of the head was impossible due to the pain associated with it; there was also no evidence of the patient turning her head while sleeping. The sternocleidomastoid muscle was not prominent and the clavicle was intact. The neurological status was normal. Since the symptoms had lasted for 1 week, AARF was suspected. The condition justified early plain radiographs, but as a result of poor cooperation, plain radiographs were not available. Static CT and conventional static MRI scans were performed and rotational positioning without an increased distance between the dens and the arc of C1 was determined, suggesting Fielding type 1 AARF (Figs. 1 and 2). There was no indication of trauma and no vertebral anomalies were observed. Aiming at expedited diagnosis and treatment, dynamic CT was first considered as a diagnostic procedure and also during manipulation. However, it was not

Table I. A literature review of studies published since 1998 regarding imaging modalities and treatment methods for AARF in pediatric patients.

Author (year)	Cases (n)	Study design	Patient age (years)	Imaging methods used	Treatment	(Refs.)
Beier <i>et al</i> (2012)	40	9-year retrospective		3D-CT and MRI	Rigid cervical collar. There was alar ligament damage	(18)
Mihara <i>et al</i> (2001)	35		Mean 6.5	N/A	Halter traction 2-3 weeks, 25.7% recurrence	(19)
Pang and Li (2005)	29	Prospective		Dynamic CT		(14)
Subach <i>et al</i> (1998)	20	7-year retrospective	Mean 6.4	Plain radiographs + dynamic CT	Rigid collar (n=5) and traction (n=15)	(15)
Mezue <i>et al</i> (2002)	13	2-year retrospective case series	3-12	X-ray, CT, MRI	Neck collar, NSAIDs, rest, halter traction for AARF (n=3)	(20)
Ciftedemir (2012)	12	Retrospective case series		X-ray and axial CT	Traction treatment, 3 weeks collar. All recovered	(21)
Deichmueller and Welkoborsky (2010)	12	Retrospective case series	Mean 7.1	X-ray, ultrasound, CT, MRI	4 required closed reduction and external fixation with Halo-vest, 8 had a spontaneous reduction	(22)
Tauchi <i>et al</i> (2011)	7			3D-CT	Halo-vest	(4)
Lee <i>et al</i> (2002)	2 (+4 chronic)	5-year retrospective case series	Mean 9 (7-12)	X-ray, CT	Non-operative care	(23)
Rahimi <i>et al</i> (2003)	6 (AARF) (of total 23)	12-year retrospective case series	Mean 7.5 (1.8-14.6)	X-ray, dynamic CT	Immobilization with collar, traction	(24)
Been <i>et al</i> (2007)	2 (of total 4)	Case series	6.5	CT, 3D-CT	Spontaneous reduction of benign torticollis	(25)
Fernández Cornejo (2003)	4	7-year retrospective		CT + 3D-CT	Non-operative care	(26)
Holcomb <i>et al</i> (2001)	4	2-year retrospective case series	N/A	N/A	N/A	(27)
Martínez-Lage <i>et al</i> (2001)	4	5-year retrospective	Mean 8.2	X-ray and CT of all, 3D-CT of 3, MRI of 1		(28)
Ortiz <i>et al</i> (2013)	3	Case series	N/A	X-ray, CT, MRI	NSAIDs, immobilization, traction/surgery	(29)
Muniz and Belfer (1999)	2	Case series	N/A	N/A	Non-operative	(30)
Meek <i>et al</i> (2001)	1	Case report	4	Plain radiographs	Non-operative care for 1 week	(31)
Galer <i>et al</i> (2005)	1	Case report	3 (female)	Dynamic CT	Non-operative	(32)
Sobolewski <i>et al</i> (2008)	1	Case report	5 (male)	Dynamic CT	Manual reduction under sedation	(33)
Pilge <i>et al</i> (2011)	1	Case report	11 (female)	X-ray, CT	Manual reduction under GA, cervical collar for 2 weeks + antibiotics	(34)

Table I. Continued.

Author (year)	Cases (n)	Study design	Patient age (years)	Imaging methods used	Treatment	(Refs.)
Wurm <i>et al</i> (2004)	1	Case report	3 (male; 5 days of symptoms)	CT and MRI	Manual reduction under GA, antibiotics for upper respiratory tract infection, Minerva	(35)
Missori <i>et al</i> (2005)	1	Case report	7 (female)	X-ray, CT, MRI	Cervical brace	(36)
Maile and Slongo (2007)	1	Case report	3 (male)	3D-CT	24 h skeletal traction, followed by 3 weeks of cervical soft brace	(16)
Gourin <i>et al</i> (2002)	1	Case report	7 (male)	X-ray, CT, MRI	Halo-traction	(37)
Park <i>et al</i> (2013)	1	Case report	9 (male)	Dynamic CT, Fielding 2, static MRI	Neck brace, halter traction, 14-day follow-up with dynamic CT and MRI	(38)
Sia <i>et al</i> (2012)	1	Case report	7 (male)	X-ray, CT, CT	Cervical collar, medicines, traction 2 weeks later	(39)
Barcelos <i>et al</i> (2014)	1	Case report	7 (1 weeks' torticollis)	CT	Soft cervical collar	(17)
Tsai and Chou (2005)	1	Case report	4 (female; 1 week after fall)	3D-CT	Manipulation under GA	(40)

AARF, acute atlantoaxial rotatory fixation; 3D, three-dimensional; CT, computed tomography; MRI, magnetic resonance imaging; GA, general anesthesia; NSAIDs, non-steroidal anti-inflammatory drugs; N/A, information not available.

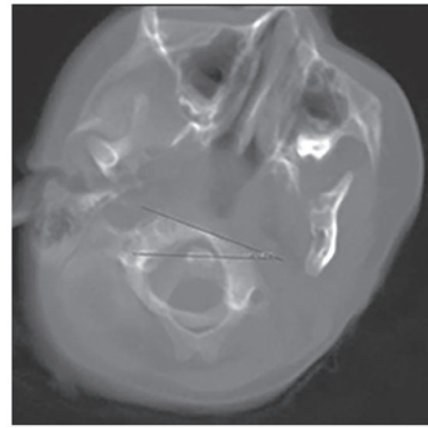


Figure 1. Two-dimensional reconstruction of CT (layer thickness, ~20 mm) of a C1-C2 region of a female patient aged 4 years with neck pain and head tilted to the left for 1 week. The CT images present an abnormal association between the C1 and C2 vertebrae matching Fielding type 1 atlantoaxial rotatory fixation. The rotational angle is 19 degrees, but the atlantoaxial distance is <3 mm. CT, computed tomography.

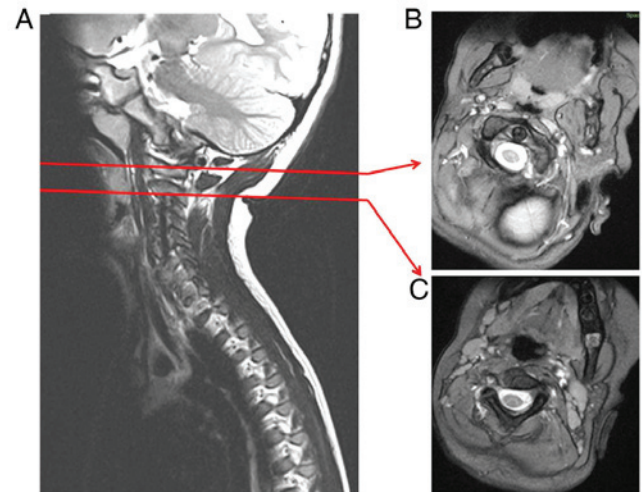


Figure 2. A female patient aged 4 years suffered acute neck pain and her head was turned to the left. Normal, conventional static magnetic resonance images of the upper cervical spine prior to manipulation were captured. (A) Sagittal image of the patient's neck and spine with the red lines depicting the precise location of the axial images in association with the sagittal image. (B) Axial projection at the C1 level. (C) Axial projection at the C2 level.

considered optimal due to the ionizing nature of the radiation and lack of soft-tissue identification. Therefore, a new study protocol (termed as interventional MRI for AARF in the present study) was set up: Scanning of the upper cervical spine by means of 1.5T interventional MRI (Siemens Magnetom Espree, Siemens Healthcare GmbH, Erlangen, Germany) with a single T2-weighted sequence to ensure a fast-operating time with reliable diagnostic accuracy. General anesthesia (GA) was required due to the severe pain associated with the movement of the cervical spine. The patient's head was carefully manipulated by a pediatric orthopedic surgeon (JS) under GA, and a T2-weighted sequence was obtained with the head rotated maximally to the right. Axial traction of the head was manually maintained during the manipulation. Subsequently, the head was rotated maximally to the left in the primary malposition

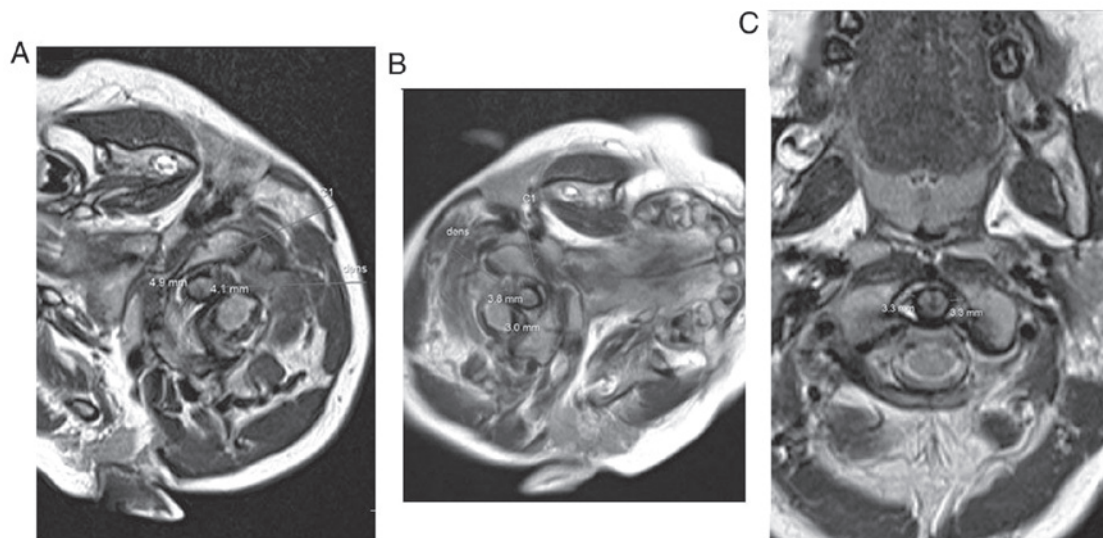


Figure 3. A 4-year-old female patient with a one-week history of neck pain and the head turned to the left was treated under general anesthesia using interventional MRI for AARF procedure. (A) Axial traction was performed manually. The orthopaedic surgeon was situated behind the head of the patient and was placing his hands bilaterally over the (mandibular) low jaw angle of the patient. The head was turned from the left to the maximal right position, during traction. The non-equal measurements in the image refer to atlantoaxial distance i.e. the distance between dens and the base of the atlas at left and right sides, which suggests AARF. (B) The patient's head was then manipulated back to the maximal left position. The atlantoaxial distance measurements were slightly unequal which indicated that a full reduction was not achieved. (C) Following return of the head to the neutral position, the measurements confirmed the symmetrical dens-atlas distances. Thereafter, the neck was immobilized with a rigid neck collar to the neutral position. The rotatory fixation was reduced during the first right-to-left manipulation and the result of treatment was controlled by simultaneous interventional MRI, instead of traditional dynamic computed tomography. MRI, magnetic resonance imaging.

direction and the interventional MRI sequence was repeated. A pediatric radiologist experienced in childhood musculoskeletal trauma (MP) reviewed all interventional MRI findings and C1-C2 fixation was determined to be reduced (Fig. 3). A clinician also had access to images to determine the immediate result of the treatment. A correct association between C1 and C2 remained stable despite movements of the head. The head was immobilized using a rigid neck collar (Aspen® Pediatric Collar, size 5; Aspen Medical Products, Irvine, CA, USA) in a neutral position and prior to awakening of the patient, one more interventional MRI scan was performed to ensure maintenance of the reduction. No dynamic CT scans were required, and clinical recovery was complete at the 6-month follow-up.

Imaging modalities in the current literature. Among the published original studies reviewed, the major modality for imaging AARF was CT, but plain radiographs, 3D-CT, dynamic CT and MRI were also used. A few comprehensive review articles were published, usually in connection with case reports, and 3D-CT and dynamic CT were regarded as the current methods (17). None of the studies reported on the use of interventional MRI as an elementary part of orthopaedic treatment, in the same manner as dynamic CT. Dynamic CT is classified as repeated static CT images during the reduction manoeuvre, with the head turned in several positions, rather than real-time CT-monitoring or CT fluoroscopy. Systematic analysis of the literature was based on 28 original clinical studies and case reports published on the subject including 207 patients in total (4,14-40). About half of the studies were retrospective patient series, mostly with just a few patients, while 13 were case reports. A comparison of the features of the studies is provided in Table I.

Discussion

A normal cervical spine allows for a great range of motion and most of the rotational torsion occurs at the atlantoaxial joint. When a patient presents with acutely diminished rotational movement, numerous different etiologies are possible, but fixed rotation of the atlantoaxial joint should be considered. Atlantoaxial subluxation remains a rare pediatric orthopedic condition, but it may result in permanent dysfunction of the C1-C2 joint, with a requirement for surgical fixation. When the condition is suspected (beyond benign torticollis), expedited diagnosis and treatment are crucial to minimize long-term sequelae (41).

Due to the complex structure of the first and second vertebrae of the cervical spine, it is challenging for radiologists and clinicians to interpret plain radiographs in acute conditions. These vertebrae are unique compared with the rest of the spinal column, and their anatomy allows for wide rotational motion. The C1 vertebra has no vertebral body *per se*. For that reason, in the case of pediatric patients with suspected AARF, CT is the current means to investigate potential unilateral facet subluxation and anterior displacement between the arch of C1 and the dens (42). However, given that dynamic CT with ionizing radiation exposure and poor soft-tissue resolution is not optimal in young pediatric patients, interventional MRI was used in the present study as the primary imaging modality in the orthopaedic treatment of a patient with low-grade AARF to monitor reduction performed under GA. The interventional MRI method was proven to be feasible for investigating AARF and ensuring that reduction is achieved during the procedure. This orthopedic technique, performed using interventional MRI, was further analyzed in the light of the published literature. A systematic and comprehensive literature search was performed

to determine the current means of practice in AARF. The literature search was performed using an appropriate search strategy in two different international databases: PubMed and Google Scholar. The reference lists of all relevant articles were reviewed in order to identify any studies not identified in the above searches. In the studies finally included, a considerable number of patients ($n=207$) suffering the rare condition were analyzed. Several different investigation methods were reported in these studies. CT and/or dynamic CT were used in 24 out of 28 studies and it is also the recommended method in certain handbooks (43). On the other hand, conventional MRI was used for evaluating AARF in several articles included in this literature review ($n=9/28$ of the articles; Table I). However, none of these previous studies appeared to use interventional MRI as a method for testing and assessment of reduction.

The present study was based on a radiation-free intervention for treating AARF in pediatric patients. A limitation of the present study was that only one case was reported and not a large cohort of patients. There was no plan for a prospective trial and no prior ethics board approval was obtained. However, the procedure was considered to be safer than conventional dynamic CT and therefore, no ethics board approval was required. Avoiding ionizing radiation is preferred, if alternative methods are available. A considerable amount of radiation exposure is averted when MRI is used instead of CT. Diagnostic CT was performed initially to exclude bone fractures; closed manual reduction of concomitant AARF is not recommended in fracture cases, but open surgical fixation is favored. The present study does not suggest using interventional MRI in treating syndromic patients due to potential anaesthetical challenges, with the shorter treatment time of dynamic CT preferred. Furthermore, the treatment was scheduled as urgent, as delayed reduction may lead to poor outcomes, and timely treatment was considered necessary. The present study reported on an encouraging approach for low-stage AARF treatment with interventional MRI demonstrated to be fit for purpose. The present findings may lead to future improvements in the care of pediatric patients suffering from acute AARF.

In conclusion, the present study demonstrated the feasibility of interventional MRI in AARF treatment for a pediatric patient. The preliminary results merit further investigation to evaluate interventional MRI prior to it being considered the method of choice for AARF diagnostics and treatment. Therefore, a larger study involving randomization of patients between dynamic CT and interventional MRI may be required.

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Availability of data and materials

The datasets, including closer clinical characteristics used during the present study, are available from the corresponding author on reasonable request.

Authors' contributions

JH and JS drafted, wrote and revised the manuscript for submission. MP, NS, WS and RBS drafted and revised the manuscript. All authors have contributed in the study protocol and to the initiative in reporting the issue. All authors read and approved the final manuscript.

Ethical approval and consent to participate

Not applicable.

Patient consent for publication

The legal guardian of the patient provided informed consent for the publication of this didactic case with the patient not identifiable.

Competing interests

The authors declare that they have no competing interests.

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