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Case Report

Rare giant complex composite odontoma of mandible in mixed dentition: Case report with 3-year follow-up and literature review

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ABSTRACT

Introduction: and importance: Complex odontomas are hamartomas representing a conglomeration of all dental tissues. When developed, they rarely become giant and even more rare to become giant in children. This report presented an unusual giant mandibular complex odontoma in very young patient. It also provided a literature analysis and better understanding of clinical features of such giant lesions in the mandible.

Case presentation: An 8-year-old boy presented with swelling in the right side of the mandible. After clinical and radiological exams, a provisional diagnosis of giant composite odontoma was made. This lesion led to jaw expansion, asymmetrical face, and teeth eruption alterations. Surgical excision of the lesion was performed, and histopathological examination confirmed the diagnosis. The patient's follow-up showed that normal jaw size and facial symmetry was restored with no evidence of recurrence for 3 years.

Clinical discussion: A literature review of reported mandibular large odontomas was made. All published reported cases of giant odontoma in the mandible caused bone expansion with or without pain. According to the literature, this paper described the youngest patient with giant complex odontoma in the mandible. This was also the first reported case from Syria.

Conclusion: Giant complex composite odontoma of the mandible can develop at any age causing facial asymmetry, however, with good prognosis and predictable surgical treatment.

1. Introduction

A hamartoma is a non-neoplastic disorganized proliferation of cells and tissues that are usually found in the organ from which they arise [1]. Odontomas develop as a result of hamartomatous proliferation of the dental apparatus [2]. These have been also called "composite odontomas", because they are composed of more than one type of tissues [3]. They can be classified according to the degree of similarity to normal teeth into compound and complex odontomas [4]. Compound odontomas tend to occur as multiple tooth-like structures between teeth, while complex odontoma is usually a conglomerate mass of dentin, enamel and cementum [5]. Complex odontomas are less common than compound odontomas [6]. Small odontomas can surgically be accessed and removed without difficulty, unlike large odontomas that may be problematic [7]. Luckily, odontomas can rarely exceed the size of a tooth in the regions where they develop [8]. Complex odontoma with a diameter of more than 30mm can be termed "a giant complex odontoma" [4]. Hereby, this report aims to describe an unusual case of giant complex odontoma in the mandible of young boy. It has been reported in line with the SCARE criteria [9]. Moreover, a literature search in PubMed, and Google Scholar (from January 2010 to July 2021) has been performed with the combination of keywords: (mandible OR mandibular) AND (large OR giant) AND (complex odontoma). Accordingly, a literature review describing cases of large odontomas in the mandible has been detailed.

2. Case presentation

An 8-year-old male was referred to the Department of Oral and Maxillofacial Surgery at Damascus hospital (Damascus, Syria), with a considerable swelling in the right side of his lower face. This was a result of buccal cortical bone plates expansion in the right mandible. The alveolar ridge was also swollen in the lingual and crestal/occlusal directions (Fig. 1a). This made him complained biting on the overlying

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mucosa when occluding. The swelling was hard and nontender by extra- and intraoral palpating, and caused obvious facial asymmetry (Fig. 1b). Extraoral examination also revealed no visible or palpable lymph nodes. The patient complained no pain and a slight limitation of mouth opening. Medical history revealed no significant health issue, and no history of trauma was reported. Intraorally, right mandibular primary first and second molars were present, while permanent molars in that side of the mandible were not (Fig. 1a). Buccal and lingual alveolar bone enlargement was clear with normal intact covering mucosa, except for some shallow impressions of the opposite occlusion. The bone swelling extended posteriorly from the mandibular primary second molar region towards the ramus.

A panoramic X-ray image showed a large cauliflower-like radiopaque mass with well-defined radiolucent rims occupying the entire posterior right region of the body of the mandible (Fig. 2a). Compared to the left side of the mandible, this mass had interrupted eruption of the right permanent first molar, interfered with the supposed position of the right second premolar bud, caused resorption of the right primary second molar roots, and absence of the right permanent second molar bud (Fig. 2a). It also extended exceptionally towards the inferior border of the mandible yielding to asymmetrical inferior displacement of the border. The mandibular canal was also displaced inferiorly. Computed tomography (CT) image revealed the lobular nature of this mass (Fig. 2b). According to the measurements done on the CT image, it measures 44mm in its larger diameter. Expansion of intact, but very thin, buccal and lingual cortical bone was obviously shown on the CT (Fig. 2b).

The lesion was surgically exposed and completely excised under general anesthesia and extraoral approach (Fig. 1c). The buccal bone plate was thin enough to be penetrated with a chisel only. The exposed mass was encapsulated, lobulated and had rough surface with many tiny shiny enamel pearls. The permanent first molar bud was tightly bound to the lesion capsule, so it had to be removed during the excision. The cavity was cleaned and heavily irrigated with saline. The surgical wound was closed layer by layer using absorbable suture. Skin was eventually sutured with 5-0 monofilament polyamide (nylon). Postoperative medication included antibiotic and non-steroidal anti-inflammatory drug. The patient was discharged 4 hours after surgery without intermaxillary fixation, but kept on soft food for a month. The excisional biopsy was sent for histopathological examination.

No neoplastic activity was observed, and the diagnosis of complex odontoma was confirmed by the histopathologist. Skin sutures were

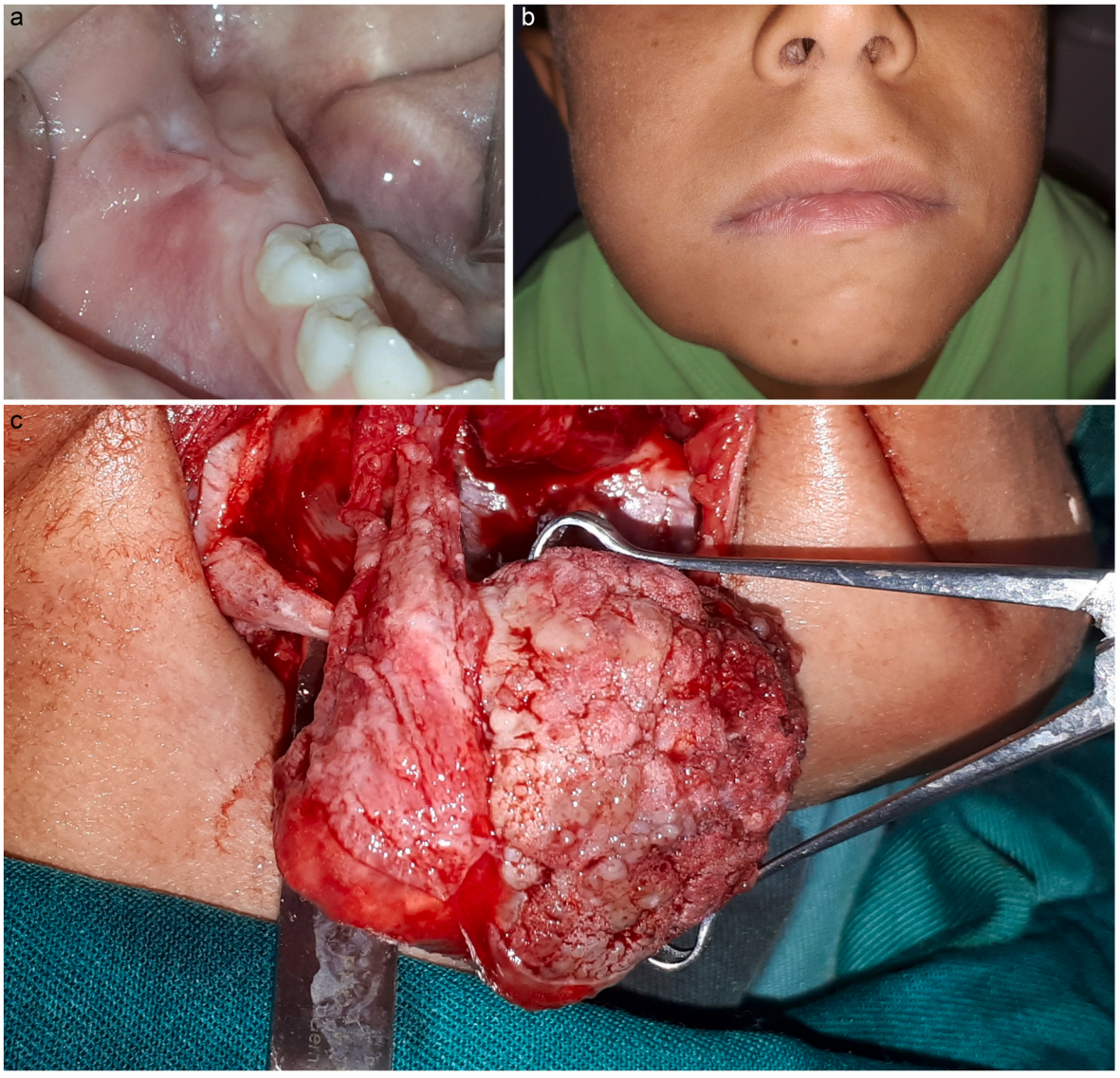

Fig. 1. Intraoral examination of the patient showed bone expansion in the right side of the mandible with noted impressions of opposite teeth on the covering mucosa (a); Jaw expansion caused obvious facial asymmetry (b); Surgical excision of the lesion showing the lobular rough surface, and the attached lesion capsule (c).

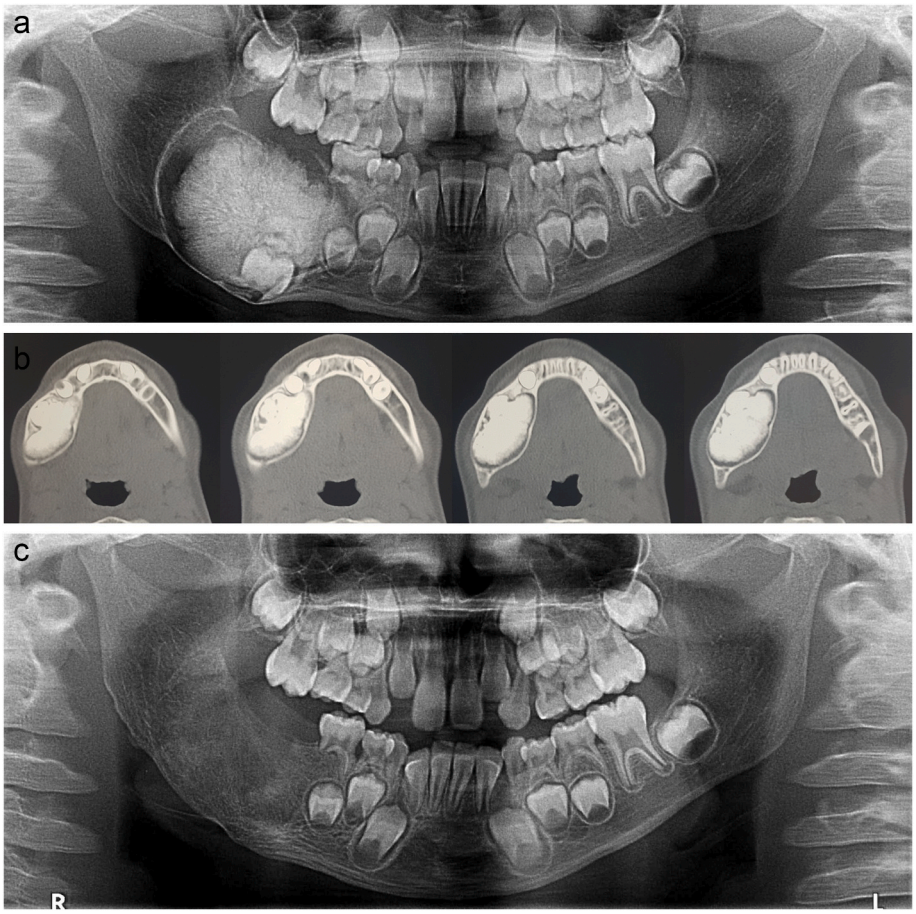

Fig. 2. Large radiopaque lesion with clear radiolucent borders occupying the entire height of the mandibular body, displacing the mandibular inferior border and mandibular canal inferiorly, extending from the right premolars region towards the right ramus, and interfering with teeth eruption (a); Patient computed tomography (CT) images showing the lobular nature of the lesion, and clear expansion of buccal and lingual bone plates (b); Favorable bone healing was seen via 6-month follow-up panoramic x-ray (c).

removed 10 days post-surgery. The patient was followed-up for 6 months and showed uneventful healing. Furthermore, facial symmetry was recovered. The patient did not report any complication or compliant. Fine-line surgical scar was noted. Favorable bone healing with no evidence of recurrence was presented by the 6-month and 3-year follow-up dental panoramic radiographs (Fig. 2c; Fig. 3).

3. Discussion

Odontomas are dental-apparatus-related hamartomas [2]. Tooth eruption alterations are the most significant characteristics of odontomas [10]. Most of odontomas do not cause clinical symptoms, and many are detected incidentally or after investigating the reason for delayed eruption of a permanent tooth [10,11]. Odontomas are frequently capable of inhibiting eruption of neighboring teeth [5]. Complex odontoma was also reported to cause devitalization of adjacent erupted teeth [12]. Limited mouth opening was reported as unusual finding associated with complex odontoma [13]. An et al. see that complex odontomas are not related to age [6]. However, they are usually seen in the second and third decades of life, and rarely seen below 10 years of age [14]. In this case report, an unusual giant complex odontoma was presented in a child of 8 years old only. According to the literature, this mandibular giant complex odontoma is the case with the youngest reported age. This is also the first reported giant complex odontoma from Syria.

On reviewing cases of giant complex odontoma of mandible in the literature, we have found that only 15 cases were reported heretofore [4,8,15–27] (Table 1). Seven patients (46.7%) were females, and eight (53.3%) were males. Their mean age was 23.13 ± 12.25 years old. The size of these large odontomas ranged from 29 to 60 mm with a mean

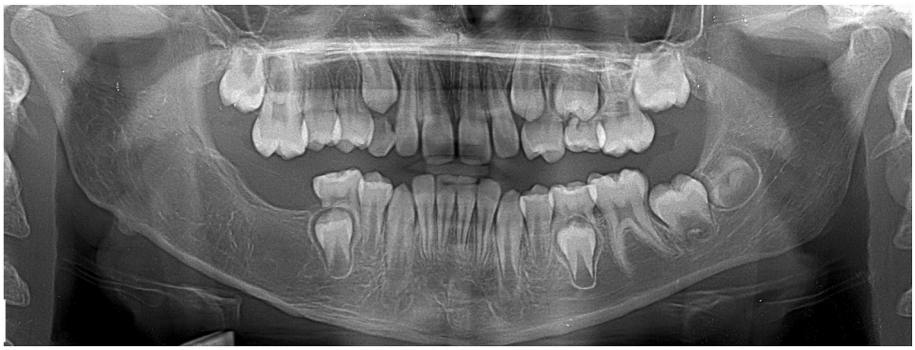

Fig. 3. Patient's dental panoramic x-ray 3 years after surgery showing no evidence of recurrence. Bilateral symmetrical mandible was restored.

Table 1
Description of giant complex odontomas of the mandible reported in the literature.

Study, Location	Gender	Age	Diameter	Pain	Bone Expansion	Facial Asymmetry	Description
Balaji and Balaji 2021, Tamil Nadu (India)	Female	24	33 mm	Painful	+	NR	Large complex composite odontoma in the right mandibular posterior region associated with cervical lymphadenopathy, first molar impaction and compression of the inferior alveolar nerve [15].
Aschaitrakool et al., 2021, Chiang Mai (Thailand)	Female	19	40 mm	Painless	+	+	Large complex odontoma of the right mandible associated with inferior displacement of the mandibular right third molar and the inferior alveolar canal [16].
Bueno et al., 2020, Sao-Paulo (Brazil)	Male	42	NA	Painless	+	+	Giant complex odontoma in the left mandibular angle extended towards the ramus and the distal side of the first molar, and associated with local infection and limited mouth opening [17].
Botelho et al., 2019, Almada (Portugal)	Female	53	31 mm	Painful	+	NR	Multiple complex odontoma in the right side of the mandible associated with mesially displaced first premolar [18].
Saravanan et al., 2019, Tamil Nadu (India)	Female	12	40 mm	Painless	+	+	Large complex odontoma in the left angle of the mandible [19].
Park et al., 2018, Iksan (Korea)	Female	28	30 mm	Painful	+	+	Giant complex odontoma in the right side of the mandible associated with an impacted third molar [4].
Akerzoul et al., 2017, Rabat (Morocco)	Male	35	NA	Painless	+	+	Giant complex odontoma in the position of left mandibular third molar associated with inferior displacement of the mandibular canal [20].
Widayanti et al., 2017, Bandung (Indonesia)	Female	17	NA	Painless	+	+	Extensive complex odontoma in the left side of the mandible associated with teeth impaction, thinning and expansion of mandibular cortical bone [21].
Bagewadi et al., 2015, Uttar Pradesh (India)	Male	22	40 mm	Intermittent Pain	+	+	Giant complex odontoma in the region of right mandibular posterior teeth erupted partially into the oral cavity and associated with inferior displacement of mandibular canal and impaction of second premolar [22].
Reddy et al., 2014, Andhra Pradesh (India)	Male	13	29 mm	Tender	+	+	Large complex odontoma in the right mandibular angle region associated with impacted mandibular second molar [23].
Perumal et al., 2013, Limpopo (South Africa)	Female	24	55 mm	Painful	+	+	Large sequestrating complex odontoma in the right side of the mandible associated with cellulitis and purulent discharge [8].
D'Cruz et al., 2013, Bangalore (India)	Male	18	45 mm	Painless	+	+	Large complex odontoma in the left posterior mandible associated with missing first and second left molars [24].
Spini et al., 2012, Itumbiara (Brazil)	Male	9	60 mm	Painless	+	NR	Giant complex odontoma in the anterior region of the mandible associated with impaction of all anterior permanent teeth [25].
Chrcanovic et al., 2010, Belo Horizonte (Brazil)	Male	21	40 mm	Painful	+	+	Large well-circumscribed complex odontoma in the right mandibular angle associated with displacement of second molar to the mandibular base [26].
Biocic et al., 2010, Zagreb (Croatia)	Male	10	50 mm	Painful	+	+	Large erupting complex odontoma in the right side of the mandible associated with a dentigerous cyst [27].

Diameter = the largest reported diameter measured in millimeters; NA = not accurate; NR = not reported.

diameter of 41.08 ± 9.94 mm. The largest giant complex odontomas were reported in the literature by Widayanti et al. and Bueno et al. [17,21]. They presented extensive lesions of 80 mm in diameter [17,21]. However, we believe the measurements were not accurate enough. Most of mandibular giant odontomas were found in the posterior region (93.3%). 33.3% were reported in the left posterior region, 60% in the right posterior, while only one case (6.7%) was reported in the anterior mandible. Moreover, multiple odontomas occurring in more than one region of subject's jaws were reviewed by Botelho et al. [18]. It seems that giant complex odontomas have great tendency to expand the jaw, where all reported giant complex odontomas (100%) were associated with buccal and/or lingual bone plates expansion, and most of these cases (80%) reported correlated facial asymmetry. In the maxilla, giant complex odontoma was reported not only to expand buccal and lingual plates but also to cause elevation of maxillary sinus [28]. In the presented case, cortical bone expansion occurred in all directions, i.e. buccal, lingual, crestal and inferior directions. The alveolus was swollen by the lesion causing biting of overlying oral mucosa. The inferior border of the mandible was also swollen causing significant facial asymmetry. Pain or tenderness was reported in eight cases (53.3%), while the rest (46.7%) were painless.

The etiology of giant odontoma remains unclear. Trauma, infection, and inflammatory process have been suggested as possible factors contributing to the development of odontomas [29]. Odontomas can also be inherited [30]. Moreover, genetic mutations may lead to the formation of these lesions [29,31]. As this case was presented during the Syrian crisis, exposure to chemicals and warfare-related agents might be a potential factor in the occurrence of the lesion.

Surgical removal of giant complex odontomas is the treatment of choice. One-stage as well as two-stage surgery was described in the literature [17,26]. Very extensive multiple lesions were suggested to be managed by partial surgical excision [32]. Sagittal split osteotomy of the mandible has been proposed as an access to complex odontomas in the mandible to reduce morbidity [33]. There was a publication reported managing giant complex odontoma with hemmimandibulectomy [21]. This radical excision, even with thinning and expansion of mandibular bony plates, was unjustified. Based on the literature and this report, more conservative surgeries were very predictable [4,22,23,25]. Piezoelectric technology could be used in removing the external cortical bone and exposing the lesion to avoid potential damage to the inferior alveolar nerve [17]. Some surgeons preferred to remove the large lesions by blocks to reduce morbidity [17,27]. However, one-piece surgical removal of the giant composite odontoma was done in the present case to ensure the complete removal of the lesion and its fibrous capsule. This was mainly facilitated by the extraoral surgical approach.

Histopathological confirmation of odontoma diagnosis is very important. Unfortunately, this is not a routine practice in real life with such cases [34]. Deferential diagnosis of complex odontoma include a wide range of lesions that have radiopaque mass surrounded by radiolucent borders [5]. Complex odontoma can radiographically resemble ossifying fibroma, osteoblastoma, and osteoma [35]. Odontomas were reported to be associated with other odontogenic lesions, like

adenomatoid odontogenic tumor, and more commonly with dentigerous cysts [27,35,36]. The prognosis of odontomas is, in general, very favorable with very rare tendency towards recurrence [10]. However, if recurrence occurs, one should think of other lesions, e.g., ameloblastic fibro-odontoma and calcifying odontogenic cyst [5].

Although panoramic x-ray has many known limitations [37], routine dental panoramic imaging may pose an important role in incidentally detecting these silent lesions before they become giant and symptomatic causing jaw expansion and facial asymmetry. Early detection may facilitate early surgical removal of these odontomas with minimal morbidity and complications that include loss of permanent teeth as unluckily happened in this case. Fortunately, this patient did not complain neurological complications after surgery despite the large lesion size and the associated inferior displacement of the mandibular canal. However, eliminating such potential risk is not warranted with giant complex odontoma [38].

4. Conclusion

This report presented an unusual giant mandibular complex odontoma in very young patient, and provided a literature analysis and better understanding of clinical features of such giant lesions in the mandible. Giant complex odontoma can occur at any age with no gender predilection, can be associated with pain or tenderness, and almost always causes bone expansion and facial asymmetry. It generally has good prognosis, and careful surgical excision is predictable treatment.

Ethical approval

The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. Institutional ethical approval was not required.

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Author contribution

Each named author has substantially contributed to conducting this research and drafting this manuscript. All authors assume full responsibility for its content. All those who have made substantive contributions to the article have been named as authors. Authors contributions were as follows: Conceptualization, N.S., N.M.A., M.A.; Methodology, N.S., N.M.A., M.A.; Data curation, N.S. and N.M.A.; Formal analysis, N.M.A.; Writing- original draft, N.S. and N.M.A.; Writing-review and editing, N.M.A. and M.A.; Final approval, N.S., N.M.A., M.A.

Registration of research studies

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Guarantor

All authors accept full responsibility for the work and conduct of the study, had access to the data, and approved the decision to publish this work in *Annals of Medicine and Surgery*.

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Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Provenance and peer review

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Declaration of competing interest

Each named author has no conflict of interest, financial or otherwise.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.amsu.2022.103355.

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