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Enchondromas of Long Bones and Other Skeletal Lesions Found Incidentally Need Critical Evaluation, But Rarely Systematic Follow-Up

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Abstract

Purpose: Incidental bone lesions are a challenge for the specialist, who has to give recommendations for further management. This review of our cases will assist in the decision whether the lesion can be "neglected", needs further active follow-up or direct initiation of treatment. **Patients and Methods:** 153 cases of incidental bone findings were presented to our musculoskeletal tumor service for evaluation from July 2008 through June 2021. 73 of them were cartilaginous tumors and 63 of these were diagnosed as enchondroma of a long bone based on X-Ray and MRI. **Results:** Follow-up imaging of the enchondroma patients was available for 35 patients at 1 to 13 years (mean 4.3 y), with no change in size except for one femoral diaphyseal enchondroma with increasing diameter from age 18 to 20 years. 14 additional patients answered written contact stating that they remained asymptomatic at 2 to 12 years (mean 5.6 y). None of the patients has been reported to the Swiss Confoederation Cancer Registry to have developed malignancy. Among the 10 other cartilaginous tumors were one chondrosarcoma grade II exhibiting different imaging, 3 non-long-bone localizations (pelvis, scapula and rib), 2 Ollier-type enchondromas, and 2 osteochondromas. Incidental findings other than cartilaginous tumors were fibrous dysplasia (n = 31), non-ossifying fibroma (n = 31) and 18 other "sporadic" entities. **Conclusions:** Incidentally found enchondromas not exhibiting aggressive features need no systematic follow-up and patients can be "discharged" with the advice to present, if symptoms would develop. This also applies to fibrous dysplasia

and the other sporadic lesions. 6 cases with other diagnoses needed specific treatment.

Keywords

Incidental Bone Lesions, Enchondroma, Fibrous Dysplasia, Non-Ossifying Fibroma

1. Introduction

Each case presenting with an incidental finding in musculoskeletal imaging raises concern regarding diagnosis and further management.

Among the series of incidental bone findings referred to our tumor group for evaluation cartilaginous tumors were the most frequent, usually referred with the objective to "rule out sarcoma".

Several authors (Patel *et al.* [1], Davies *et al.* [2], and Ahmed *et al.* [3]) recently questioned, whether follow-up imaging is needed in incidentally found painless chondroid lesions in long bones without aggressive features in imaging.

Historically the risk of developing chondrosarcoma ex enchondroma has been overestimated, as enchondromas were detected in standard radiographs only in the presence of distinct calcifications. In the era of MRI, enchondromas are found more frequently, and with a prevalence of 2.8% in knee MRI according to Stomp *et al.* [4] may be regarded as a "normal concurrent finding".

With our data, we wish to contribute to the discussion of how to proceed with enchondromas found incidentally regarding follow-up. We also include the other incidental bone lesions to expand on differential diagnostic aspects.

2. Materials and Methods

Patients

153 cases have been referred by orthopaedic surgeons to our dedicated musculoskeletal tumor group for the evaluation of incidental bone findings from July 2008 through June 2021. Imaging was performed in most patients because of shoulder or knee problems. All patients had standard X-Ray and MRI documentation. The diagnoses as an incidental finding were based upon the patients' history and physical examination and are summarized in **Table 1**.

Based on X-Ray and MRI 73 of the patients had cartilaginous tumors and 63 of these were diagnosed as enchondroma of a long bone. One of the 63 had bilateral enchondromas in the distal femur. Histologic examinations were performed on 5 of the 63 enchondroma cases. One humeral lesion was curetted elsewhere for suspected chondrosarcoma and eventually diagnosed as enchondroma without atypia; one patient with a distal tibia enchondroma had a biopsy as an external radiology expert felt another entity needed to be ruled out.

Table 1. List of incidentally found bone lesions regarding diagnosis, size of the enchondromas, management and follow-up data.

Incidental Bone Lesions						
Localization	n	Age Range (Mean)	Maximum diameter Range (Mean)	Cortical contact (n) Scalloping	Management	Follow-up Range (Mean)
Enchondroma long bones n = 63, *for illustration see* **Figures 1-3**						
Humerus proximal	18	38 - 77 y (53y)	1.0 - 8.0 cm (4.5 cm)	Scalloping <1/3 in 2, >2/3 in 2 no change during 3 y f/u Other patients no scalloping	1 biopsied at arthroscopy 1 curetted to exclude sarcoma; Histology on both "enchondroma"	Imaging for 12 patients available at 1 - 7 y (3.69 y) no change Clinically asymptomatic 4 additional patients at 4, 9, 10, 12 y
Humerus diaphysis	2	42 - 53 y	1.5/9.0 cm	No scalloping		1 patient asymptomatic at 12 y
Femur distal	27	34 - 72 y (50.8 y)	0.8 - 11.0 cm (3.5 cm)	Only the patient with the 11 cm long enchondroma had minimal scalloping remaining unchanged at 7 y f/u	2 cases curetted and filled with graft to prepare for cruciate ligament reconstruction/knee replacement Histology: enchondroma	Imaging for 9 patients available at 1 - 7 y (3.4 y) no change 8 additional patients asymptomatic at 2 - 10 y (5.6 y)
Femur mid-diaphysis	4	18 - 34 y (26.5 y)	2.7 - 4.6 cm (3.3 cm)	No scalloping		1 patient followed from age 18 y to age 20 y showed increase in diameter from 3.2 to 4.6 cm without scalloping—to be followed
Femur proximal	2	62/51 y	7.0/2.3 cm	No cortical contact		Imaging for 1 patient at 2 y—no change
Tibia proximal	4	22 - 55 y (45 y)	2.1 - 6.8 cm (4.0 cm)	1 mild scalloping eccentric lesion unchanged over 4 y f/u		Imaging available for 3 patients at 3, 4, 8 y—No change
Tibia distal	1	53 y	3.0 cm	Eccentric with extracortical extrusion	Biopsy: benign enchondroma	No f/u

Continued

Fibula proximal	5	43 - 59 y (52 y)	1.0 - 4.5 cm (2.9 cm)	All patients showed some scalloping in the fibular head	1 curetted, bone plasty enchondroma	Imaging available for 6 patients at 3 - 13 y (7.4 y) no change; 2 additional patient asymptomatic at 3, 12 y
Other cartilaginous lesions n = 10						
Diagnosis/ Localization	n	Age	Remarks		Treatment	
Chondrosarcoma grade II proximal Tibia	1	30 y	**Figure 4**		Wide resection, endoprosthesis	
Enchondroma type Ollier Femur distal	1	53 y			Observation	
Enchondroma type Ollier Tibia distal	1	34 y			Observation	
Osteochondroma Fibula	3	21 - 56 y (42 y)			Resection	
Periosteal chondroma Femur distal	1	34 y			Wide resection	
Chondrosarcoma exostotic grade I Scapula	1	19 y			Wide resection	At f/u 4 y no recurrence
Myxoid chondrosarcoma Pelvis	1	31 y			Wide resection	
Enchondroma Rib	1	35 y			Observation	At 2 y f/u unchanged
Fibrous dysplasia including LSMFT, for illustration see **Figure 5**						
Femur 13, Pelvis 7, Rib 1, Ulna 1, Fibula 2, Tibia 7	31	11 - 72 y (43 y)			4 cases biopsy proven	
Non-ossifying fibroma/fibrous cortical defect, for illustration see **Figure 6**						
NOF/Fibrous histiocytoma	31	3 - 52 y (22 y)			2 cases curetted and bone plasty for diagnostic purposes	

Continued

Other incidental findings n = 18				
Cortical desmoid Femur kondyle	4	10 - 24 y (16.8 y)	Reference [26]	Observation
Osteofibrous dysplasia with rhabdoid elements	1		Reference Exner [29]	Spontaneous regression
Simple juvenile bone cyst Humerus	1	4 y		Observation
Simple juvenile bone cyst distal Fibula	1	18 y		Observation
Periosteal vascular malformation distal Femur	1	57 y		Biopsy/observation
Calcaneus Cysts/Lipomas	7	12 - 71 y (35 y)	Reference Malghem [28]	1 filled with DBF at same time as resection of fibula osteochondroma; 2 "prophylactically" filled with DBF
Osteopoikily	3		No long bones affected	

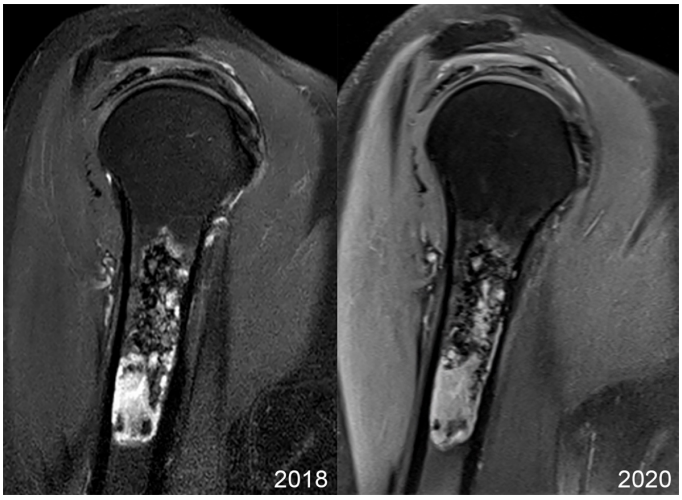

Figure 1. Female, 60 years: Enchondroma. MRI for rotator cuff evaluation. Proton-density-fat-saturated images show the enchondroma with a length of 63 mm unchanged over 2.8 years.

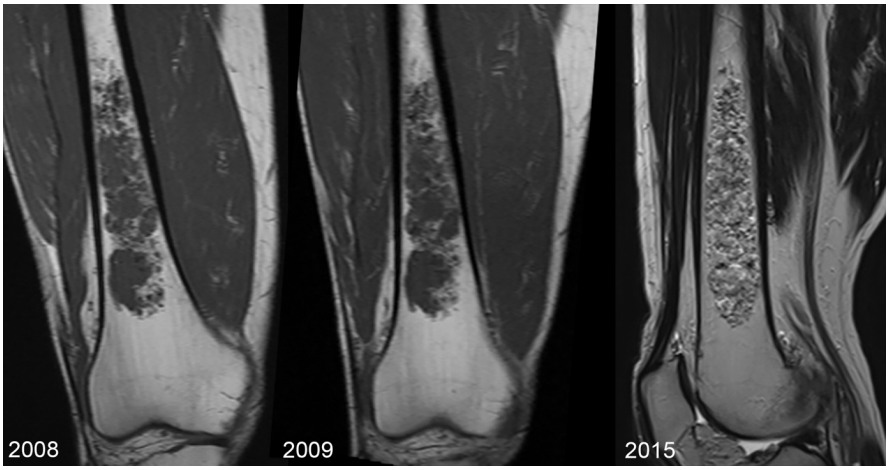

Figure 2. Male, 63 years: Enchondroma. MRI for evaluation of meniscal problems. The length (126 mm) and structural characteristics of "rings and arcs" remained unchanged over 7 years. In 2015 only sagittal images were provided. Clinically the patient is asymptomatic in 2021.

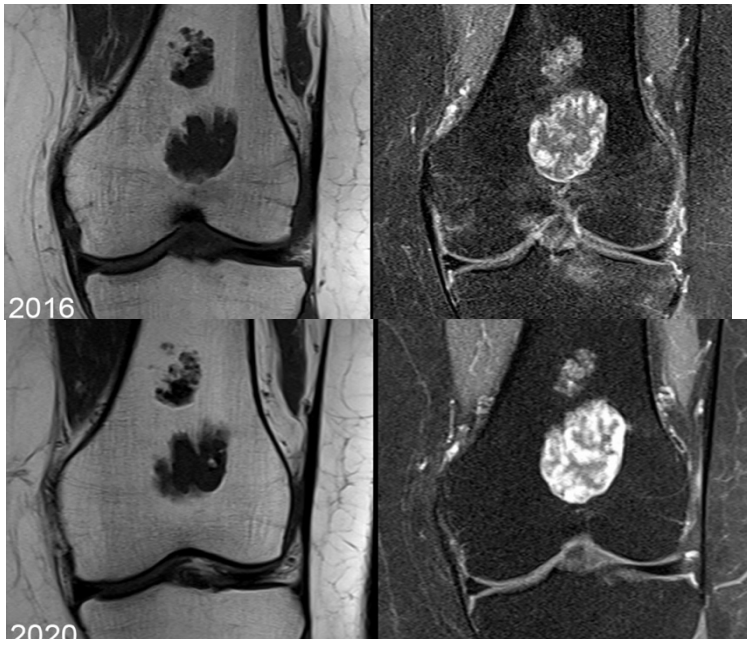

Figure 3. Female, 50 years: Enchondroma. Knee MRI for evaluation after contusion of the knee joint. The presentation of two separate lesions is unusual, but the images are typical for a chondroid matrix with inhomogenous contrast enhancement in the T1 weighted fat saturated images. No increase in size over the 4.5 years follow-up (top 2016, bottom 2020).

Two enchondroma cases of the femur found incidentally were curetted and filled with bone to prepare for later knee replacement or cruciate ligament reconstruction respectively; one proximal fibula enchondroma was curetted elsewhere.

One 30-year-old patient with an atypical radiological finding in the MRI performed for suspected ligament injury from distorsion of the knee joint (**Figure 4**) was eventually diagnosed as chondrosarcoma grade II of the medial tibial pla-

teau and treated by wide resection and endoprosthetic reconstruction.

It is mandatory to report any malignant tumor including atypical chondromatous lesions to the Swiss Confoederation Cancer Registry. None of the patients of this study has been recorded up to January 2022, thus further documenting uneventful development.

Other cartilaginous lesions considered to be enchondromas type Ollier (n = 3), and osteochondromatous lesions are listed for completeness.

Follow-up with imaging had been recommended to the referring colleagues at first presentation. Request for follow-up information was written at the time of this study. Patients were also asked for information about their present health.

3. Results

Follow-up imaging is available for 35 of the 63 enchondroma patients at 1 to 13 years (mean 4.3 years) with no change in size, except for one femoral diaphyseal enchondroma with increasing diameter from 3.4 mm at age 18 years to 4.2 mm at age 20 years. 14 patients answering to personal contact by phone or letter responded to have remained asymptomatic at follow-up over 2 to 12 years (mean 5.6 years). 14 patients could not be contacted because of having moved to an unknown address.

The data of the enchondroma and non-enchondroma lesions are summarized in Table 1. MRIs of representative cases of enchondroma, chondrosarcoma, fibrous dysplasia and non-ossifying fibromas are presented in Figures 1-6.

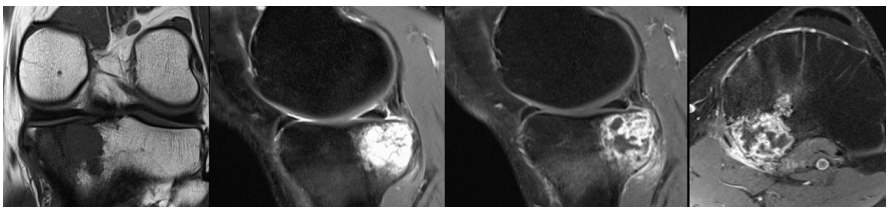

Figure 4. Male, 31 years: Chondrosarcoma grade II. MRI performed for suspected meniscal lesion after distorsion of the knee joint. The pattern is consistent with a ring-and-arc chondroid matrix. However, it exhibits aggressive features such as endosteal scalloping and soft tissue extension. Furthermore, the intraepiphyseal location must raise suspicion for an atypical process.

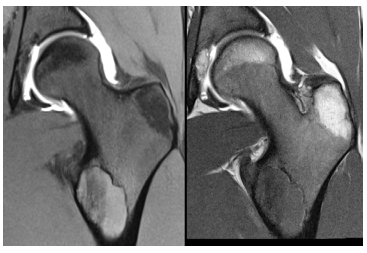

Figure 5. Female, 23 years: Fibrous dysplasia. Arthro-MRI was performed for suspected labral lesion. Localized defect in osteoblastic differentiation and maturation with replacement of normal bone by large fibrous stroma. Coronal proton density weighted image with fat saturation (left) showing higher signal compared to the T1 weighted image (right). Physiologic signal in the femoral head and greater trochanter.

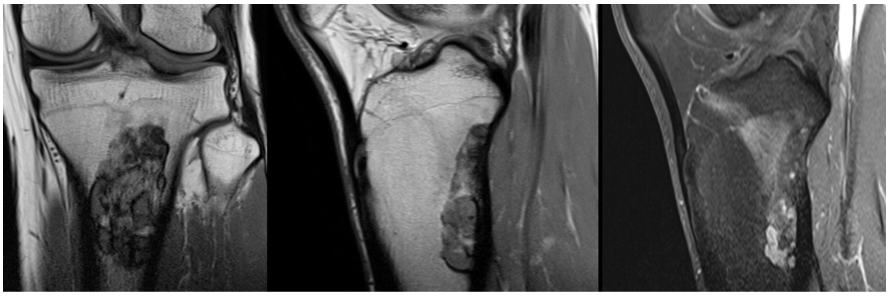

Figure 6. Male, 19 years: Large non-ossifying fibroma/fibrous cortical defect. This lesion already "matured" with ossification and consequently low signal on all sequences; T1 (a); proton density weighted (b); T1 fat-saturated and contrast enhancement (c).

4. Discussion

The purpose of this study is to assist in the decision making when faced with an incidental long bone finding presented to a musculoskeletal specialist.

Extensive literature upon the question of how to manage incidental orthopaedic findings exists; we, therefore, cite only a few selected references (Kim *et al.* [5], Velasco *et al.* [6], Nguyen *et al.*, [7], Hiremath *et al.* [8], du Preez *et al.* [9]). Vanel *et al.* [10], Notrott *et al.* [11]).

Until recently follow-up imaging of incidentally found enchondromas has been generally considered mandatory. Since this strategy has been challenged by Ahmed *et al.* [3], we wish to contribute to this question with our data. In addition, we want to shed some light on other incidental bone findings.

Diagnoses encountered in our cases have been as follows.

4.1. Enchondroma

Tumors producing a chondroid matrix are histologically classified as cartilaginous neoplasms (Fletcher *et al.* [12]) and comprise a spectrum ranging from resting lesions to highly aggressive malignant neoplasms. Enchondroma, periosteal chondroma, enchondromatosis (Ollier and Maffucci Syndrome) and osteochondroma ("exostosis") are benign tumors grouped together as chondromas, pathologically sharing hyaline cartilage. Chondroblastoma, chondromyxoid fibroma and synovial chondromatosis also exhibit cartilaginous differentiation.

Chondrosarcomas are malignant cartilage-differentiating tumors graded usually II or III.

Enchondromas are benign cartilaginous neoplasms that develop within the medullary space of bone and are derived from growth plate chondrocytes in bone (Milgram *et al.* [13], Zhang *et al.* [14]). Enchondromatous lesions continuing to grow and exhibiting destructive features have historically been reported as low-grade chondrosarcoma, grade 1. However, as they do not metastasize and therefore don't fulfill criteria for malignancy, recently they are reported as central atypical cartilaginous tumor and should no longer bear the label "sarcoma" (WHO Classification of Tumours [12].

The differentiation between enchondroma and central atypical cartilaginous tumor hinges on imaging features as the histology may be identical (Nguyen *et al.* [7]). Aggressive features suggesting chondrosarcoma include endosteal scalloping more than two third of length of the lesion, large non-calcified component, and larger lesions (Murphey *et al.* [15], Choi *et al.* [16]). Choi *et al.* [16] in their study of 18 patients with low-grade chondrosarcoma and 16 patients with enchondroma, both groups with histo-pathological examinations, found higher incidence of predominantly intermediate signal in T1-weighted images, multilocular appearance on contrast-enhanced T1-weighted images, cortical destruction, soft tissue mass and adjacent abnormal bone marrow and soft tissue signal among the low-grade chondrosarcoma cases.

Scalloping is seen in most cases of proximal fibula and humeral head enchondromas, which may be explained by the thin cortex, and in these locations not necessarily indicate aggressiveness.

However, there are no clear-cut imaging criteria to separate enchondroma from low-grade chondrosarcoma with a high interobserver inconsistency regarding diagnosis and grading (Geirnaerdt *et al.* [17]), Jones *et al.* [18], Gelderblom *et al.* [19], Deckers *et al.* [20], Eefting *et al.* [21]).

Endosteal scalloping was found to allow differentiation between enchondroma and chondrosarcoma when involving more than one-third of the lesion (Murphey *et al.* [15]). Longitudinal tumor extent more than 5 cm was found to be a predictable indicator of malignancy (Geirnaerdt *et al.* [17]).

However, the data indicate a low risk for the evolution from quiescent enchondroma to a more active atypical chondromatous lesion (a nomenclature now replacing the description as low grade (I) chondrosarcoma). They are therefore actually considered as "leave alone lesions"; patients should be informed about the benign nature and to watch development and present again, when the incidental lesion becomes symptomatic.

The data furthermore do not support the recommendation of curettage of enchondromas and atypical chondromatous tumors still frequently advocated (e.g. Deckers *et al.* [22]) which is associated naturally with a certain rate of complications.

If control of the incidental lesion by repeat imaging is recommended this should be at long intervals, e.g. 3 to 5 years. First, this would save expenses and secondly this warrants that the patient does not get "tired" from follow-up before changes can be expected.

The few lesions found larger than 5 cm longitudinal extent in our patients even in the presence of scalloping showed no progression within the time frame of observation (**Figures 1-3**).

4.2. Other Incidentally Found Lesions

Osteochondromas are readily diagnosed and except for cases with a large cartilaginous cap can be resected without further preoperative investigations (WHO

classification of Tumours [12]). The terminology "(osteo-)cartilaginous exostoses" is not any longer recommended (WHO classification of Tumours [12]).

Fibrous dysplasia typically presents an incidental finding in its monostotic form. Follow-up consultation is needed only when symptoms develop (DiCaprio *et al.* [23]).

The liposclerosing myxofibrous tumor (LSMFT) is a benign fibro-osseous lesion, has distinct radiographic features and is generally diagnosed incidentally (Deel *et al.* [24]). Heim *et al.* [25] proposed that LSMFT probably represents a traumatized variant of fibrous dysplasia and involutional/ischemic changes can be so extensive that residual fibrous dysplasia areas are completely absent.

Non-ossifying fibromas are easily diagnosed and rarely need intervention, but apparently, even orthopaedic surgeons may have difficulties to make the diagnosis especially in larger lesions (Herget *et al.* [26]).

The so-called periosteal desmoids are mentioned as we consider them normal variants of no relevance except for ruling out neoplasms mimicking these minor abnormalities (Tscholl *et al.* [27]).

Clinical relevance of calcaneal cysts depends upon size (Pogoda *et al.* [28]). It is of interest that calcaneal cysts and calcaneal lipomas may be a single entity and a pathogeneic continuity may exist (Malghem *et al.* [29]).

The incidentally found case of the osteofibrous dysplasia with rhabdoid elements and spontaneous resolution has been published recently (Exner *et al.* [30]).

5. Drawbacks and Limitations of This Study

Follow-up of the patients in our observational group is incomplete and with an average of 4.1 years short for lesions, which only at very long term, probably decades, may change their activity.

Patients not followed by the authors usually stay in the initial medical setting; therefore they most likely would have presented to the referring orthopaedist or to our group in case of symptoms or imaging changes of their bone lesion.

Furthermore, the diagnoses were almost exclusively based on imaging findings alone.

Other lesions that typically present incidentally as e.g. intraosseous hemangioma were not encountered in this series but need to be recognized.

6. Summary and Conclusions

Our findings agree with those of Ahmed *et al.* [3], Patel *et al.* [1], and Davies *et al.* [29] that asymptomatic patients with long bone enchondromas less than 5 cm longitudinal extension with no endosteal scalloping do not need systematic imaging follow-up and can be discharged after explanation of the diagnosis and instruction to seek medical attention in case of any change, development of discomfort or increasing pain.

The lesions larger than 5 cm have remained stable in our patients; however,

follow-up is too short to exclude progression into chondrosarcoma grade II or even grade III at longer intervals. Therefore we cannot provide data on the time interval needed to develop dedifferentiation. Until more data are available it may therefore be cautious to repeat imaging. This, however, should be done at rather longer time intervals; 5-year intervals may be reasonable as with shorter intervals patients not seeing definitive progression would become reluctant to submit to further examinations creating unnecessary concerns and last but not least costs. We hope that systematic follow-up data will add to the scientifically based care of patients with incidentally found enchondromas.

Ethical Approval

The study was approved by the institutional review boards.

Informed Consent

Informed consent was obtained from all patients.

Authors' Contribution

The first author collected the data, all authors contributed equally to the evaluation and interpretation of images, and finalizing the manuscript.

Conflicts of Interest

There are no conflicts of interest.

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