



# Assessment of radiological and clinical follow-up strategies for intrathoracic osteochondromas in hereditary multiple osteochondromas patients

Niyazi İğde, MD¹, Osman Emre Aycan, MD², Berkay Doğan, MD¹, Berksu Polat, MD¹, Serhat Gürbüz, MD¹,

<sup>1</sup>Department of Orthopedics and Traumatology, Baltalimani Bone Diseases Training and Research Hospital, İstanbul, Türkiye <sup>2</sup>Department of Orthopedics and Traumatology, University of Health Sciences, Hamidiye Faculty of Medicine, İstanbul, Türkiye

Hereditary multiple osteochondromas (HMOs) is an autosomal dominant skeletal disorder characterized by the development of multiple osteochondromas.[1] These benign bone tumors typically arise in the metaphyseal regions of long bones where active bone growth occurs; however, they may also be observed in other skeletal structures such as the ribs, spine, and pelvis.[1-7] The diagnosis is usually established radiologically by demonstrating two or more originating from osteochondromas regions. [5] Due to their locations, large sizes, and multiplicity, osteochondromas can lead to various health issues and may negatively affect overall health status.[6]

Received: July 28, 2025 Accepted: September 18, 2025 Published online: December 29, 2025

Correspondence: Niyazi İğde, MD. Baltalimani Kemik Hastalıkları Eğitim ve Araştırma Hastanesi, Ortopedi ve Travmatoloji Kliniği, 34470 Sarıyer, İstanbul, Türkiye.

E-mail: niyazi.igde@gmail.com Doi: 10.52312/jdrs.2026.2522

Citation: İğde N, Aycan OE, Doğan B, Polat B, Gürbüz S. Assessment of radiological and clinical follow-up strategies for intrathoracic osteochondromas in hereditary multiple osteochondromas patients. Jt Dis Relat Surg 2026;37(x):i-xi. doi: 10.52312/jdrs.2026.2522.

©2026 All right reserved by the Turkish Joint Diseases Foundation

This is an open access article under the terms of the Creative Commons Attribution-NonCommercial License, which permits use, distribution and reproduction in any medium, provided the original work is properly cited and is not used for commercial purposes (http://creativecommons.org/licenses/by-nc/4.0/).

#### **ABSTRACT**

**Objectives:** This study aims to evaluate the incidence, distribution, clinical implications, and surgical criteria for intrathoracic osteochondromas in hereditary multiple osteochondroma (HMO) patients.

Patients and methods: Between January 2010 and January 2024, a total of 246 patients (146 males, 100 females; mean age: 8.2±4.5 years; range, 1 to 18 years) who were diagnosed with HMO and had thoracic imaging using computed tomography (CT) with at least one year of clinical follow-up were retrospectively analyzed. Lesion characteristics (type, size, number, and location), symptoms, and surgical data were recorded. The receiver operating characteristic (ROC) curve analysis was performed to identify the size threshold associated with surgical indication.

**Results:** Intrathoracic osteochondromas were detected in 35% of chest X-rays and in 81.8% of CT scans. Among 45 CT-confirmed cases, 68.9% had multiple lesions and 77.8% were pedunculated. Surgical excision was performed in 12 patients (4.9%), most commonly due to chest pain or paracardiac location. Tumor size was significantly higher in excised cases (median: 26 mm *vs.* 12 mm, p<0.001). A lesion size >15 mm predicted surgical need with 91.67% sensitivity and 72.73% specificity (area under the curve [AUC]=0.883, p<0.001).

**Conclusion:** Routine chest radiography, reinforced by selective thoracic CT in symptomatic or suspicious cases, should be integrated into the follow-up of HMO patients to detect intrathoracic osteochondromas early. A lesion size of >15 mm, chest pain, and paracardiac localization are strong predictors of surgical intervention and should guide clinical decision-making.

**Keywords:** Hereditary multiple osteochondroma, imaging studies, intrathoracic osteochondroma, surgical indications.

Intrathoracic osteochondromas are typically asymptomatic and often detected incidentally; however, they have the potential to cause serious complications such as pneumothorax,

ii Jt Dis Relat Surg

hemothorax, cardiac compression, and pulmonary contusion. [8-11] Although rare, these complications may be life-threatening and require early diagnosis and appropriate management. [2-12] Nevertheless, current knowledge regarding intrathoracic osteochondromas is predominantly limited to case reports, and large-scale studies systematically evaluating their incidence, anatomical distribution, and clinical features in patients with HMO are lacking. [2,12-14] Additionally, aspects such as the growth rate of thoracic osteochondromas, their clinical progression, and the role of lesion size in determining surgical indications remain insufficiently clarified.

Most studies in the literature addressing HMO-related lesions primarily focus on extremity deformities, joint limitations, or spinal involvement, while intrathoracic lesions have been relatively underexplored. [4,7] To the best of our knowledge, no large case series has systematically evaluated the frequency, anatomical distribution, and clinical characteristics of intrathoracic lesions in patients with HMO. [8-11]

In the present study, we hypothesized that routine chest radiography, with selective use of computed tomography (CT) in symptomatic or radiographically suspicious cases, would allow early detection of intrathoracic osteochondromas in patients with HMO, and that lesion characteristics such as size, paracardia clocalization, and associated symptoms could reliably predict the necessity of surgical intervention. We, therefore, aimed to assess the incidence, anatomical distribution, associated complications, and surgical indications of intrathoracic osteochondromas in patients with HMO and to emphasize the importance of detailed thoracic evaluation during routine follow-up of these patients, to facilitate early recognition of potential complications, and to contribute to the clarification of radiological and clinical criteria used in surgical decision-making.

# **PATIENTS AND METHODS**

This single-center, retrospective study was conducted at Baltalimani Bone Diseases Training and Research Hospital, Department of Orthopedics and Traumatology between January 1st, 2010 and January 1st, 2024. Initially, a total of 371 patients with HMOs were screened. Of these patients, 246 pediatric patients (146 males, 100 females; mean age: 8.2±4.5 years; range, 1 to 18 years) who underwent posteroanterior (PA) chest radiography and had regular clinical follow-up for at least one

year were included. Among these patients, 55 had available thoracic CT imaging. Patients who did not complete follow-up, missed scheduled visits, or did not undergo thoracic imaging were excluded. Written informed consent was obtained from the parents and/or legal guardians of the patients. The study protocol was approved by the Baltalimani Kemik Hastalıkları Eğitim ve Araştırma Hastanesi (Date: 28.10.2024, No: 194). The study was conducted in accordance with the principles of the Declaration of Helsinki.

Clinical and radiological parameters were analyzed in detail. First, the patients' age and sex distribution were evaluated, and findings or complications that developed during follow-up were systematically recorded. The type of osteochondroma (sessile or pedunculated) was identified, and the size, location, and number of lesions were analyzed. Additionally, local complications such as pain, pulmonary parenchymal irritation, hemothorax, and cardiac compression were assessed.

### **Imaging methods**

In our clinic, radiography (X-ray) was primarily used for the diagnosis and follow-up of patients. Computed tomography was performed in patients presenting with thoracic symptoms, such as chest pain or respiratory complaints. In some cases, CT was requested due to suspicion of an intrathoracic lesion on chest X-ray. Additionally, patients with a diagnosis of HMO, who underwent thoracic CT for unrelated reasons were also evaluated. Magnetic resonance imaging (MRI) is a crucial imaging modality in the surveillance and evaluation of osteochondromas, particularly in cases with suspected malignant transformation, due to its lack of ionizing radiation and its superior capability in accurately assessing cartilage cap thickness.<sup>[15]</sup> However, thoracic MRI is subject to limitations due to motion artifacts arising from respiratory and cardiac movements, which often compromise image quality.[16] This challenge is further pronounced in the pediatric population, where the need for sedation and extended scanning durations represent an additional limitation.[17] In our institution, the inability to perform sedation during MRI examinations further restricts its routine applicability. Conversely, the potential carcinogenic effects of repeated exposure to ionizing radiation represent a significant concern, particularly in children. To reduce this risk, we used low-dose CT protocols adjusted for age and body size. [18] The literature reports that such techniques can

reduce radiation dose by approximately 50 to 80% while maintaining diagnostic image quality. [19] For lesion size assessment, CT imaging was utilized, and all measurements were conducted using the Extreme PACS software. In patients with multiple osteochondromas, the largest lesion was selected for measurement. In pedunculated lesions, the size was measured from the base of the stalk to the most distal point of the mass, whereas in sessile lesions, the maximum diameter was recorded.

# Surgical indications and follow-up protocol

In our clinic, HMO patients are evaluated from the time of diagnosis with follow-up visits scheduled every six months or annually. At the time of initial diagnosis, all patients undergo two-view radiographs of the four extremities, pelvis, and chest. The MRI is requested for lesions identified on these radiographs. During follow-up, imaging (X-ray, MRI, or CT) is planned based on patient complaints and physical examination findings, particularly for palpable or symptomatic lesions. Computed tomography is used when clinically indicated, particularly in cases with suspected intrathoracic involvement or complications. Intrathoracic lesions that are large or associated with complications are referred to thoracic surgery. While some patients undergo surgical intervention, others are jointly followed by orthopedic and thoracic surgery teams without the need for surgery.

TABLE I			
Demographic and clinical features and hemithoracic osteochone	dromas dis	tribution	in HMO cases
	n	%	Mean±SD
Numerical variables			
Age at initial presentation (year)	246		8.2±4.5
Follow-up duration (mo)	246		73.4±55.0
Age at first CT (year)	45		10±5.5
Age at second CT (year)	7		11.2±2.9
Time interval between first and second CT (year)	7		1.6±0.8
Tumor size on first CT (mm)	45		16.7±10.6
Tumor size on second CT (mm)	7		21.1±11.0
Difference in tumor size between first and second CT (mm)	7		4.4±3.7
Categorical variables			
Sex			
Female	100	40.7	
Type of osteochondroma (due to CT)			
Pedunculated	35	77.8	
Sessile	10	22.2	
Thoracic osteochondroma (due to CT)			
Solitary	14	31.1	
Multiple	31	68.9	
Regular follow-up interval (mo)			
6	54	28.5	
12	136	71.5	
Excision of thoracic osteochondroma (present)	12	4.9	
Paracardiac osteochondromas (relative to ribs) (present)	18	40	
Regular follow-up (present)	190	77.2	
Presence of extrathoracic surgery (present)	148	60.2	
CT imaging performed (present)	55	22.4	
Mass detected on CT (present)	45	81.8	
Mass detected on PA chest X-ray (present)	86	35.0	
Chest pain (present)	12	26.7	
HMO: Hereditary multiple osteochondroma; SD: Standard deviation; CT: Compute	ed tomograpl	ny.	

iv Jt Dis Relat Surg

## Statistical analysis

Statistical analysis was performed using the SPSS version 27.0 software (IBM Corp., Armonk, NY, USA) and MedCalc 14 (MedCalc Software, Ostend, Belgium). The normality of data distribution was assessed using the Kolmogorov-Smirnov and Shapiro-Wilk-Francia tests. Continuous data were expressed in mean  $\pm$  standard deviation (SD) or median (min-max), while categorical data were expressed in number and frequency. To compare two independent groups based on quantitative variables, the independent samples t-test and the Mann-Whitney U test were used. For categorical variable comparisons, Pearson chi-square and Fisher-Freeman-Halton tests were analyzed. Sensitivity and specificity for classification based on cut-off values were assessed using the receiver operating characteristic (ROC) curve analysis. A p value of <0.05 was considered statistically significant with 95% confidence interval (CI).

## **RESULTS**

Among 246 patients included in the study, a total of 114 had intrathoracic masses. While chest masses were detected in 86 patients on PA chest radiographs, an additional 28 patients who had no visible lesions on radiographs were found to have masses on thoracic CT. Overall, thoracic CT revealed masses in 45 patients, whereas no masses were observed in the CT scans of 10 patients. The mean follow-up was 73.4±55.0 (range, 12 to 168) months (Table I).

The mean age at the first CT scan (n=45) was  $10\pm5.5$  years. For patients who underwent control CT scan (n=7), the mean age was  $11.2\pm2.9$  years. The mean interval between the first and second CT scans was  $1.6\pm0.8$  (range, 1 to 3) years (Table I).

In the first CT scan, the mean tumor size was  $16.7\pm10.6$  (range, 5 to 50) mm in 45 patients. In the second CT scan, the mean tumor size increased to  $21.1\pm11.0$  (range, 13 to 43) mm in seven patients.

TABLE II		
Hemithoracic osteochondromas distribution in HMO patients		
	n	%
Hemithoracic osteochondromas location		
Right	5	11.1
Left	21	46.7
Bilateral	19	42.2
Location of osteochondromas in left hemithorax (based on rib number) (n=40)		
1	5	7.2
2	7	10.1
3	15	21.7
4	25	36.2
5	9	13.0
6	5	7.2
8	1	1.4
10	1	1.4
12	1	1.4
Location of osteochondromas in right hemithorax (based on rib number) (n=24)		
1	3	8.6
2	5	14.3
3	8	22.9
4	3	8.6
5	1	2.9
6	3	8.6
7	6	17.1
8	3	8.6
9	3	8.6
HMO: Hereditary multiple osteochondroma.		

Among these seven patients, who had both scans, the mean increase in tumor size between the two scans was  $4.4\pm3.7$  (range, 0 to 11) mm (Table I).

Based on osteochondroma type, 77.8% had pedunculated lesions, while 22.2% had sessile osteochondromas. In terms of thoracic osteochondromas distribution, 31.1% were solitary, and 68.9% were multiple (Table I).

Among the patients under regular follow-up, 28% attended follow-up visits every six months, while 72% were followed up every 12 months. Thoracic osteochondroma excision was performed in 4.9% of the cases. Paracardiac osteochondromas was identified in 40% of the patients based on thoracic CT findings. Additionally, 60.2% of the

patients previously underwent extrathoracic surgical interventions, primarily due to osteochondromas arising in frequently affected skeletal regions including the femur, tibia, pelvis, and ilium. The CT imaging was performed in 22.4% of the patients; among them, thoracic masses were identified in 81.8% of patients undergoing CT scans, compared to 34.9% on PA chest radiographs. A total of 26.7% of the patients with a mass detected on CT reported chest pain. In terms of thoracic osteochondroma location, 11.1% of patients had right-sided involvement, 46.7% had left-sided involvement, and 42.2% had bilateral involvement (Table II).

Chest pain was more frequently observed in patients who underwent thoracic osteochondroma

		TABLE III				
Comparison of th	oracic osteoc		sion by osteoch	ondroma type		
		of thoracic nondroma		Type of osteo	ochondroma	
	Absent	Present	р	Pedunculated	Sessile	p
Age at first CT (year), median (Q1-Q3)	10 (8-14)	10.5 (7.5-15)	0.794*	10 (8-13.5)	12 (5.5-15)	0.728*
Age at initial (year), median (Q1-Q3)	8 (4-11)	9 (4.5-15)	0.313*	6 (3.5-10)	4.5 (3.5-9)	0.614*
Tumor size on first CT (mm), median (Q1-Q3)	12 (8-16)	26 (16.5-36)	<0.001*	14 (10-22)	12 (5-40)	0.790*
Follow-up duration (mo), median (Q1-Q3)	68 (28-120)	34 (13.5-109)	0.454*	72 (14-128)	111 (66-135)	0.168*
Type of osteochondroma, n (%)						
Pedunculated	27 (81.8)	8 (66.7)	0.418†	-	-	
Sessile	6 (18.2)	4 (33.3)		-	-	
Thoracic osteochondromas, n (%)						
Solitary	11 (33.3)	3 (25)	0.725†	12 (34.3)	2 (20)	0.469†
Multiple	22 (66.7)	9 (75)		23 (65.7)	8 (80)	
Paracardiac osteochondromas, n (%)						
Absent	23 (69.7)	4 (33.3)	0.041†	20 (57.1)	7 (70)	0.716†
Present	10 (30.3)	8 (66.7)	4.6 (1.1-18.9)‡	15 (42.9)	3 (30)	
Mass detected on PA chest X-ray, n (%)						
Right	2 (6.1)	3 (25)	0.246†	3 (8.6)	2 (20)	0.307†
Left	16 (48.5)	5 (41.7)		18 (51.4)	3 (30)	
Bilateral	15 (45.5)	4 (33.3)		14 (40)	5 (50)	
Presence of extrathoracic surgery, n (%)						
Absent	93 (39.7)	5 (41.7)	0.999†	10 (28.6)	1 (10)	0.409†
Present	141 (60.3)	7 (58.3)		25 (71.4)	9 (90)	
Mass detected on PA chest X-ray, n (%)						
Absent	153 (65.4)	7 (58.3)	0.757†	23 (65.7)	5 (50)	0.467†
Present	81 (34.6)	5 (41.7)		12 (34.3)	5 (50)	
Chest pain, n (%)						
Absent	28 (84.8)	5 (41.7)	0.007†	26 (74.3)	7 (70)	0.999†
Present	5 (15.2)	7 (58.3)	7.8 (1.8-34.8)‡	9 (25.7)	3 (30)	

CT: Computed tomography; Q: Quartile; PA: Posteroanterior; ‡ Odss Ratio (95% Confidence interval); \* Mann-Whitney U Test (Monte Carlo); † Fisher Freeman Halton (Monte Carlo).

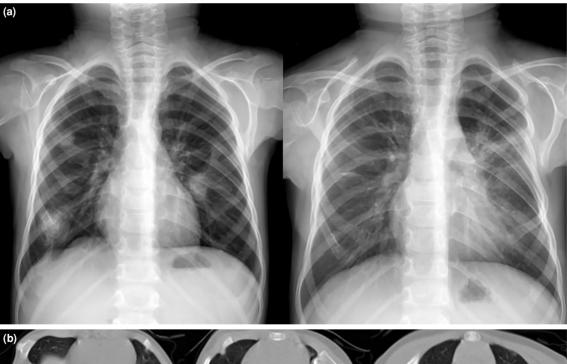
vi Jt Dis Relat Surg

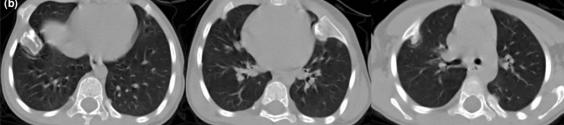
excision (p=0.007). The median tumor size on thoracic CT was significantly greater in the excision group versus those managed conservatively: 26 (range, 16.5 to 36) mm in 12 patients vs. 12 (range, 8 to 16) mm in 33 patients (p<0.001; (Table III). Furthermore, paracardiac osteochondromas was more common in excised cases (8/12, 66.7%) than in conservatively followed patients (10/33, 30.3%) (p=0.041) (Table III, Figure 1a, b, Figures 2 and 3)

Among 45 patients in whom thoracic osteochondroma was detected on thoracic CT, 31 (68.9%) had multiple and 14 (31.1%) had solitary lesions. Paracardiac osteochondromas were present in 18 (40%) patients. There was no statistically significant difference in the frequency of thoracic

or paracardiac osteochondromas between male and female patients (p>0.05). The median tumor size at the time of the first CT scan was significantly larger in patients with multiple osteochondromas 16 (range, 12 to 23) mm compared to those with solitary lesions 11.5 (range, 5 to 13) mm (p=0.021) (Table IV).

Among 45 patients who underwent thoracic CT, pedunculated-type osteochondromas were identified in 35 patients, while sessile-type lesions were observed in 10 patients. Of those with pedunculated lesions, 23 (65.7%) had multiple osteochondromas and 12 (34.3%) had solitary lesions. Among patients with sessile-type lesions, eight (80%) had multiple and two (20%) had solitary





**FIGURE 1.** (a) Preoperative (left) and postoperative 1-year (right) posteroanterior chest radiographs of a 4-year-old male patient. Large osteochondromas are visible in the right hemithorax above the diaphragm and in the left paracardiac region, both located in anatomically high-risk areas due to their size and proximity to vital structures. Thoracic CT was performed for detailed evaluation, and the lesions were subsequently excised to prevent potential complications. (b) Preoperative thoracic CT image of the same patient showing multiple intrathoracic osteochondromas. The lesions identified on PA chest radiograph were further evaluated with CT to assess their risk, and surgical excision was planned accordingly. CT: Computed tomography.



**FIGURE 2.** Thoracic CT image of a 6-year-old female patient showing a pedunculated paracardiac osteochondroma. Findings consistent with pulmonary parenchymal contusion are present.

CT: Computed tomography.

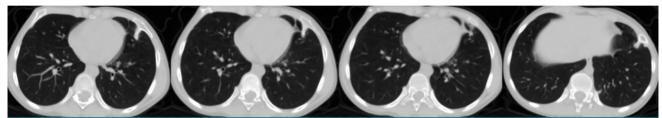


FIGURE 3. Thoracic CT image of a 12-year-old male patient demonstrating multiple pedunculated paracardiac osteochondromas. CT: Computed tomography.

osteochondromas. Of the 45 patients, 12 underwent surgical excision. Among these, nine (75%) patients had multiple osteochondromas, while three (25%) patients had solitary lesions. Although pedunculated osteochondromas were usually more common, the lesion type did not show a marked difference based on whether the osteochondromas was solitary or multiple (Table IV).

In terms of the necessity for thoracic osteochondroma excision, the sensitivity and specificity for patients with tumors larger than 15 mm on the first CT scan were 91.67% and 72.73%, respectively. The ROC analysis revealed an area under the curve (AUC) value of 0.883, indicating a statistically significant difference (p<0.001).

## **DISCUSSION**

This study represents a comprehensive evaluation focusing on the incidence, anatomical distribution, associated complications, and surgical indications of intrathoracic osteochondromas in patients with HMO. By analyzing a large cohort of 246 HMO patients, we attempt to fill the gap in the literature concerning thoracic involvement, which has been relatively under-investigated compared to spinal and extremity skeletal lesions.

The main finding of this study was the detection of costal osteochondromas in 35%

of chest radiographs (86/246 patients) and in 81.8% of thoracic CT scans (45/55 patients). Notably, 28 patients had osteochondromas that were not visible on chest radiographs, but were detected on CT imaging performed due to thoracic symptoms, and six of these patients underwent surgical excision. Furthermore, among patients who underwent a second CT scan for follow-up of previously identified intrathoracic osteochondromas, a mean increase in lesion size of 4.4±3.7 mm was observed over a mean follow-up period of 1.6±0.8 years. These findings highlight the clinical relevance of intrathoracic osteochondromas and highlight the necessity of systematic thoracic evaluation during routine follow-up to prevent serious complications.

A noteworthy observation was the association between tumor size and clinical outcomes. In previously published case reports describing hemothorax and cardiac compression, the excised lesions were reported to be approximately 3 cm in size. [20,21] Similarly, in our study, the median size of osteochondromas in patients, who underwent surgical excision, was 26 (range, 16.5 to 36) mm, which was significantly larger than those followed conservatively (median: 12 [range, 8 to 16] mm), with a statistically significant difference (p<0.001). According to ROC analysis, a tumor size >15 mm on CT demonstrated high sensitivity (91.67%) and

	TABLE IV					
Solitary and multiple thoracic osteochondromas in relation to the presence of paracardiac involvement	dromas in relation	to the presence	of paracardiac invo	olvement		
	Thoracic osteochondromas	chondromas		Paracardiac osteochondromas (relative to ribs)	eochondromas to ribs)	
	Solitary	Multiple	Q	Absent	Present	d
Age at initial presentation (year), median (Q1-Q3)	5 (4-7)	6 (3-10)	0.955*	6 (4-10.5)	4.5 (2.5-8)	0.178*
Tumor size on first CT (mm), median (Q1-Q3)	11.5 (5-13)	16 (12-23)	0.021*	12 (8-22)	16 (12-19)	0.336*
Follow-up duration (mo), median (Q1-Q3)	101 (39-128)	72 (14-135)	0.397*	100 (39-141)	52.5 (14-90)	0.068*
Type of osteochondroma						
Pedunculated	12 (85.7)	23 (74.2)		20 (74.1)	15 (83.3)	
Sessile	2 (14.3)	8 (25.8)	0.469†	7 (25.9)	3 (16.7)	0.716 <del>†</del>
Present	10 (30.3)	8 (66.7)		15 (42.9)	3 (30)	
Hemithoracic osteochondromas location (relative to ribs)			0.002			
Right	3 (21.4)	2 (6.5)	NS	4 (14.8)	1 (5.6)	
Left	10 (71.4)	11 (35.5)	0.025	12 (44.4)	6 (50)	0.828†
Bilateral	1 (7.1)	18 (58.1)	0.001	11 (40.7)	8 (44.4)	
Mass detected on PA chest X-ray						
Absent	10 (71.4)	18 (58.1)		15 (55.6)	13 (72.2)	, pr
Present	4 (28.6)	13 (41.9)		12 (44.4)	5 (27.8)	+100.0
Chest pain						
Absent	12 (85.7)	21 (67.7)	0.007	21 (77.8)	12 (66.7)	+007
Present	2 (14.3)	10 (32.3)	7.8 (1.8-34.8)	6 (22.2)	6 (33.3)	0.499
OT. Communical semiconscience Date Date Date Date Date Date Date Dat	- 11 out: 4/V/ cach * .//o.	+ .(ola 0) ota 0) + 10 ota 0	0+10H 0000000000000000000000000000000000	+ · (0]200 0+00 W	, +00T 0,000 O 140 000	Marke Contract

CT: Computed tomography; Q: Quartile; PA: Posteroanterior; ‡ Odss Ratio (95% Confidence interval); \* Mann-Whitney U Test (Monte Carlo); † Fisher Freeman Halton (Monte Carlo); ‡ Pearson Chi-Square Test (Monte Carlo);

specificity (72.73%) in predicting the need for surgical intervention. This finding emphasizes the critical role of lesion size in clinical decision-making and supports the utility of CT for accurate risk stratification due to its precise measurement capabilities.

A recent review evaluating case reports in the literature emphasized that pneumothorax and hemothorax secondary to costal osteochondromas in HMO patients, although rare, remain potentially life-threatening conditions. It stressed the importance of initial evaluation with chest radiography and thoracic CT.[22] In the same study, among 18 reviewed case reports, the most commonly reported excision sites were the right seventh (19.0%), sixth (14.3%), and eighth (14.3%) ribs.[22] In contrast, our study found that the paracardiac region was involved in 40% (n=18) of cases, with a significantly higher rate in the surgical group compared to the non-surgical group  $(66.7\% \ vs. \ 30.3\%, \ p=0.041)$ . These findings suggest that paracardiac lesions are more likely to cause cardiac or pulmonary compression symptoms, while lesions located in the right hemithorax may be more prone to complications such as pneumothorax and hemothorax.[21,22] Although severe complications such as hemothorax or cardiac compression were infrequent in our cohort, their potential severity necessitates vigilant follow-up.

In the current study, the mean age at initial presentation was 8.2±4.5 years, with a predominance of pediatric and adolescent patients, consistent with the natural course of HMO, in which osteochondromas typically arise during periods of active skeletal growth.[23] The mean follow-up duration of 73.4±55.0 months enabled robust evaluation of lesion progression and clinical outcomes. Although osteochondromas are benign, they may cause significant mechanical complications and carry a risk of malignant transformation, particularly in hereditary cases.[24,25] While primary and secondary chondrosarcomas are more frequently reported in older patients in the literature, [25-27] no cases of thoracic chondrosarcoma were observed in our study, which included only pediatric patients. This may be attributed to the lower risk of secondary chondrosarcoma arising from osteochondromas in the pediatric age group. However, this finding does not eliminate the responsibility of orthopedic specialists to remain vigilant for thoracic lesions in children with HMO and to refer patients to thoracic surgery when clinically indicated.

In our cohort, pedunculated osteochondromas (77.8%) were more frequently identified than which aligns sessile lesions (22.2%), morphological descriptions previous HMO.[7,12-14] Pedunculated lesions with narrow stalks and sharp contours have a greater tendency to cause mechanical irritation of adjacent tissues, potentially leading to symptoms such as chest pain, hemothorax, pneumothorax, and pulmonary contusion.[2,12,14] While surgical treatment is recommended for symptomatic osteochondromas, it may also be considered in asymptomatic cases with a pedunculated, spiculated morphology to prevent life-threatening complications. [2,12] The surgical excision rate of 4.9% in our study reflects a conservative approach targeting symptomatic or high-risk lesions, indicating that clinical decisions are based more on lesion characteristics than merely the presence of osteochondromas. Additionally, 60.2% of patients previously underwent surgeries unrelated to the thorax, highlighting the multisystemic nature of HMO and the need for a multidisciplinary approach.

Regarding anatomical distribution, thoracic osteochondromas in our study were bilateral in 42.2% and left-sided in 46.7% of cases, and multiple lesions (68.9%) were more common than solitary ones (31.1%). These findings emphasize the importance of comprehensive imaging of the entire thoracic cage, particularly in asymptomatic patients, to avoid overlooking lesions. Literature reports show that thoracic osteochondromas can involve various costal segments, often present in multiple forms, and that lesion characteristics such as size, location, and orientation may vary significantly among those causing complications. [2,13,20-22,24-26] Therefore, each lesion should be evaluated individually, taking into account the clinical context and lesion-specific features.

In contrast to most previous studies, which are usually limited to case reports of intrathoracic complications, our study provides a broader perspective with a large case series systematically screened for thoracic lesions.<sup>[2,13,20-22,24,25]</sup> While prior research on HMO has predominantly focused on extremity or spinal involvement,<sup>[3,4]</sup> our data underscore the utility of routine chest radiography as a screening tool and the superior sensitivity of CT in detecting and characterizing intrathoracic lesions. More importantly, CT is not recommended as a routine imaging modality for all HMO patients; rather, it should be applied selectively in cases with clinical symptoms (e.g., chest pain, respiratory

x Jt Dis Relat Surg

complaints) or suspicious radiographic findings. This selective approach minimizes unnecessary radiation exposure.

The main limitations to this study include its retrospective design, which carries an inherent risk of selection bias, particularly as not all patients underwent CT imaging. Moreover, the low incidence of severe complications may limit the generalizability of our findings to populations with varying disease severity. Future prospective studies incorporating standardized imaging protocols could provide a clearer understanding of the natural history of intrathoracic osteochondromas and help refine screening recommendations.

In conclusion, our study results highlight the importance of systematic thoracic evaluation for the detection and management of intrathoracic osteochondromas in patients with HMO. Lesion size, chest pain, and paracardiac location were identified as key factors influencing surgical decisions. Routine chest radiography should be considered the primary screening tool, while thoracic CT imaging should be reserved selectively for symptomatic patients or when radiographic suspicion arises. Incorporating this selective approach into follow-up protocols enables early detection of clinically significant lesions and timely intervention, while minimizing unnecessary radiation exposure. Taken together, our findings support a proactive screening approach to improve clinical outcomes and quality of life in patients with HMO.

**Data Sharing Statement:** The data that support the findings of this study are available from the corresponding author upon reasonable request.

**Author Contributions:** Idea/concept, design, data collection, literature review, writing the article: N.İ.; Idea/concept, analysis, control/supervision, critical review: O.E.A.; Data collection, analysis: B.D.; Data collection, literature review: B.P.; Data collection: S.G.

**Conflict of Interest:** The authors declared no conflicts of interest with respect to the authorship and/or publication of this article.

**Funding:** The authors received no financial support for the research and/or authorship of this article.

#### **REFERENCES**

- Wininger AE, Chhabra BN, Haigler RE, Hanson DS, Scott AC. The incidence of vertebral exostoses in multiple hereditary exostoses and recommendations for spinal screening. J Pediatr Orthop 2021;41:e226-31. doi: 10.1097/ BPO.00000000000001749.
- Pan R, Lu X, Wang Z, Duan L, Cao D. Hemothorax caused by costal exostosis injuring diaphragm: A case report and literature review. J Cardiothorac Surg 2022;17:230. doi: 10.1186/s13019-022-01984-7.

 Fowler J, Takayanagi A, Fiani B, Cathel A, Sarhadi KJ, Arshad M, et al. Diagnosis, management, and treatment options: A cervical spine osteochondroma meta-analysis. World Neurosurg 2021;149:215-25.e6. doi: 10.1016/j. wneu.2021.01.148.

- Monroig-Rivera C, Bockhorn L, Thornberg D, Santillan B, Rathjen KE. Prevalence of osteochondromas in the spine in patients with multiple hereditary exostoses. JB JS Open Access 2025;10:e24.00072. doi: 10.2106/JBJS. OA.24.00072.
- Hameetman L, Bovée JV, Taminiau AH, Kroon HM, Hogendoorn PC. Multiple osteochondromas: Clinicopathological and genetic spectrum and suggestions for clinical management. Hered Cancer Clin Pract 2004;2:161-73. doi: 10.1186/1897-4287-2-4-161.
- Pacifici M. Hereditary multiple exostoses: New insights into pathogenesis, clinical complications, and potential treatments. Curr Osteoporos Rep 2017;15:142-52. doi: 10.1007/s11914-017-0355-2.
- 7. Wuyts W, Schmale GA, Chansky HA, Raskind WH. Hereditary Multiple Osteochondromas. 2000 Aug 3 [updated 2020 Aug 6]. In: Adam MP, Feldman J, Mirzaa GM, Pagon RA, Wallace SE, Amemiya A, editors. GeneReviews® [Internet]. Seattle (WA): University of Washington, Seattle; 1993-2025
- Dumazet A, Launois C, Dury S, Sailhan F, Alifano M, Dewolf M, et al. Hereditary multiple exostoses of the ribs as an uncommon cause of pneumothorax: A case report. Medicine (Baltimore) 2018;97:e11894. doi: 10.1097/ MD.0000000000011894.
- 9. Chen J, Nelson S, Tzung B, Applebaum H, DeUgarte DA. Costal osteochondroma spicule associated with pleural effusion. Pediatr Surg Int 2014;30:357-9. doi: 10.1007/s00383-013-3323-7.
- Imai K, Suga Y, Nagatsuka Y, Usuda J, Ohira T, Kato H, et al. Pneumothorax caused by costal exostosis. Ann Thorac Cardiovasc Surg 2014;20:161-4. doi: 10.5761/atcs.cr.12.01955.
- 11. Smeeing DPJ, Klein WM, Dierselhuis EF, Daniels HE. Pediatric diaphragmatic hernia induced by a rib osteochondroma. Radiol Case Rep 2024;19:2498-501. doi: 10.1016/j.radcr.2024.03.039.
- Bakhshi H, Kushare I, Murphy MO, Gaynor JW, Dormans JP. Chest wall osteochondroma in children: A case series of surgical management. J Pediatr Orthop 2014;34:733-7. doi: 10.1097/BPO.0000000000000153.
- 13. Phatak SV, Kolwadkar PK, Rajderkar D. Solitary osteochondroma of rib: A case report. Indian J Radiol Imaging 2021;16:339-40.
- 14. Patel M, Bauer TW, Santoscoy T, Ilaslan H. Osteochondroma of the fifth rib resulting in recurrent hemothorax. Skeletal Radiol 2015;44:1853-6. doi: 10.1007/s00256-015-2257-7.
- Pontes ÍCM, Leão RV, Lobo CFT, Paula VT, Yamachira VS, Baptista AM, et al. Imaging of solitary and multiple osteochondromas: From head to toe - A review. Clin Imaging 2023;103:109989. doi: 10.1016/j.clinimag.2023.109989.
- Raptis CA, Ludwig DR, Hammer MM, Luna A, Broncano J, Henry TS, et al. Building blocks for thoracic MRI: Challenges, sequences, and protocol design. J Magn Reson Imaging 2019;50:682-701. doi: 10.1002/jmri.26677.
- 17. Kozak BM, Jaimes C, Kirsch J, Gee MS. MRI Techniques to decrease imaging times in children. Radiographics 2020;40:485-502. doi: 10.1148/rg.2020190112.

- Nelson TR. Practical strategies to reduce pediatric CT radiation dose. J Am Coll Radiol 2014;11:292-9. doi: 10.1016/j. jacr.2013.10.011.
- 19. Lee SH, Kim MJ, Yoon CS, Lee MJ. Radiation dose reduction with the Adaptive Statistical Iterative Reconstruction (ASIR) technique for chest CT in children: An intra-individual comparison. Eur J Radiol 2012;81:e938-43. doi: 10.1016/j. ejrad.2012.06.013.
- 20. Harrison NK, Wilkinson J, O'Donohue J, Hansell D, Sheppard MN, Goldstraw PG, et al. Osteochondroma of the rib: An unusual cause of haemothorax. Thorax 1994;49:618-9. doi: 10.1136/thx.49.6.618.
- 21. Alnassar AS. Rib osteochondroma causing cardiac compression in a pediatric patient. Int J Surg Case Rep 2021;81:105762. doi: 10.1016/j.ijscr.2021.105762.
- 22. Sheaffer K, Hampton S, Barnard E, Patel MN, Kim L, Gendreau JL. Hemothorax and pneumothorax secondary to costal involvement in hereditary multiple exostoses: A systematic review of reported cases in the literature. Cureus 2021;13:e16326. doi: 10.7759/cureus.16326.

- 23. Rueda-de-Eusebio A, Gomez-Pena S, Moreno-Casado MJ, Marquina G, Arrazola J, Crespo-Rodríguez AM. Hereditary multiple exostoses: an educational review. Insights Imaging 2025;16:46. doi: 10.1186/s13244-025-01899-6.
- 24. Tiwari C, Borkar N, Hussain N, Khubchandani N. Solitary osteochondroma of the rib: An unusual chest wall tumor in the pediatric age group. J Cancer Res Ther 2023;19:1423-5. doi: 10.4103/jcrt.jcrt\_679\_21.
- Kikuchi R, Mino N, Matsukura T, Hirai T. Resected osteochondroma of the rib in an elderly patient. Gen Thorac Cardiovasc Surg 2010;58:588-91. doi: 10.1007/s11748-009-0570-6.
- 26. Tsuda Y, Gregory JJ, Fujiwara T, Abudu S. Secondary chondrosarcoma arising from osteochondroma: Outcomes and prognostic factors. Bone Joint J 2019;101-B:1313-20. doi: 10.1302/0301-620X.101B9.BJJ-2019-0190.R1.
- 27. Gnoli M, Gambarotti M, Righi A, Staals EL, Evangelista A, Tremosini M, et al. Secondary peripheral chondrosarcoma in multiple osteochondromas: A retrospective single-institution case series. Orphanet J Rare Dis 2024;19:63. doi: 10.1186/s13023-023-03006-8.