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CASE SERIES

Spontaneous regression of solitary osteochondromas in children: An option to consider in clinical practice

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ABSTRACT

Osteochondromas are neoplasm that belong to the family of cartilaginous histogenesis tumors and represent 90% of all forms of exostoses. As most osteochondromas are asymptomatic, underdiagnosis is frequent. Symptomatic forms usually manifest before the age of 20 years, and the most common symptoms are pain and the detection of a bony mass. Herein, we report four cases of spontaneous regression of solitary osteochondromas in the light of literature. We consider that orthopedic surgeons should take into account the possibility of spontaneous regression of these tumors, before recommending surgery. Symptoms are usually mild and we recommend following these patients with X-ray and physical examination annually.

Keywords: Exostosis, osteochondroma, spontaneous regression.

Solitary osteochondromas are neoplasms that are developed during bone growth by endochondral ossification. These benign tumors belong to the family of cartilaginous histogenesis tumors and represents 90% of all forms of exostoses.[1]

A review of the current literature reveals that spontaneous regression of osteochondromas is rarely documented. In this article, we present four cases of a solitary osteochondroma resolution in the light of literature data.

CASE REPORT

Case 1- A four-year-old boy was admitted with a mass in the right knee. There was no relevant medical history or associated trauma. Physical examination revealed a hard, smooth, non-tender, non-mobile mass over the medial aspect of the distal femur. He maintained a normal range of motion in all planes. There was no neurovascular compromise. Radiological evaluation of the knee showed a broad-based sessile osteochondroma emanating from the distal femur metaphysis without any abnormal calcifications of the cartilaginous cap. It measured 19.7 mm in diameter at the base and projected 12.5 mm away from the cortex (Figure 1). Recommendation was observation with follow-up annually. At the age of seven, the tumor was undetectable neither physically nor radiologically. Repeated physical examination showed no loss of motion or neurovascular compromise.

A written informed consent was obtained from the parents and/or legal guardians of the patient.

Case 2- A 10-year-old boy presented with a lump on his left upper arm. Physical examination revealed a bony hard mass on the anterior side of the upper arm. Radiographs showed a sessile osteochondroma on the proximal humerus. It measured 30.14 mm in diameter at the base and projected 14 mm away

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from the cortex (Figure 2). The patient did not report pain with regard to the osteochondroma, although the bony prominence was palpable. There was no neurovascular compromise. Of note, the patient had a medical history of medulloblastoma, hypothyroidism, and growth retardation. Annual follow-up showed clinical and radiographic spontaneous regression of the lesion within three years.

A written informed consent was obtained from the parents and/or legal guardians of the patient.

Case 3- A 10-year-old boy, with no relevant medical history, was admitted to our orthopedic clinic for the evaluation of a mass in his left distal femur. The mass neither caused him pain nor limited his activities. Physical examination showed no tenderness to palpation over the lesion. Neurovascular examination showed no compromise to the distal extremity. A solitary sessile osteochondroma was radiographically observed on the posteromedial aspect of the distal femoral metaphysis, it measured 52 mm in diameter from the base and projected 19 mm away from the cortex (Figure 3). He was clinically and radiologically followed at 12-month intervals. An increase of the lesion size was noticed on X-ray at one-year follow-up; however, it did not produce any musculoskeletal symptoms. In the following four years, it progressively regressed spontaneously starting to shrink at the age of 12 and resolved by the age of 15.

A written informed consent was obtained from the parents and/or legal guardians of the patient.

Case 4- An 11-year-old boy presented with a lump on his right upper arm. Physical examination revealed a bony hard mass on the lateral side of the

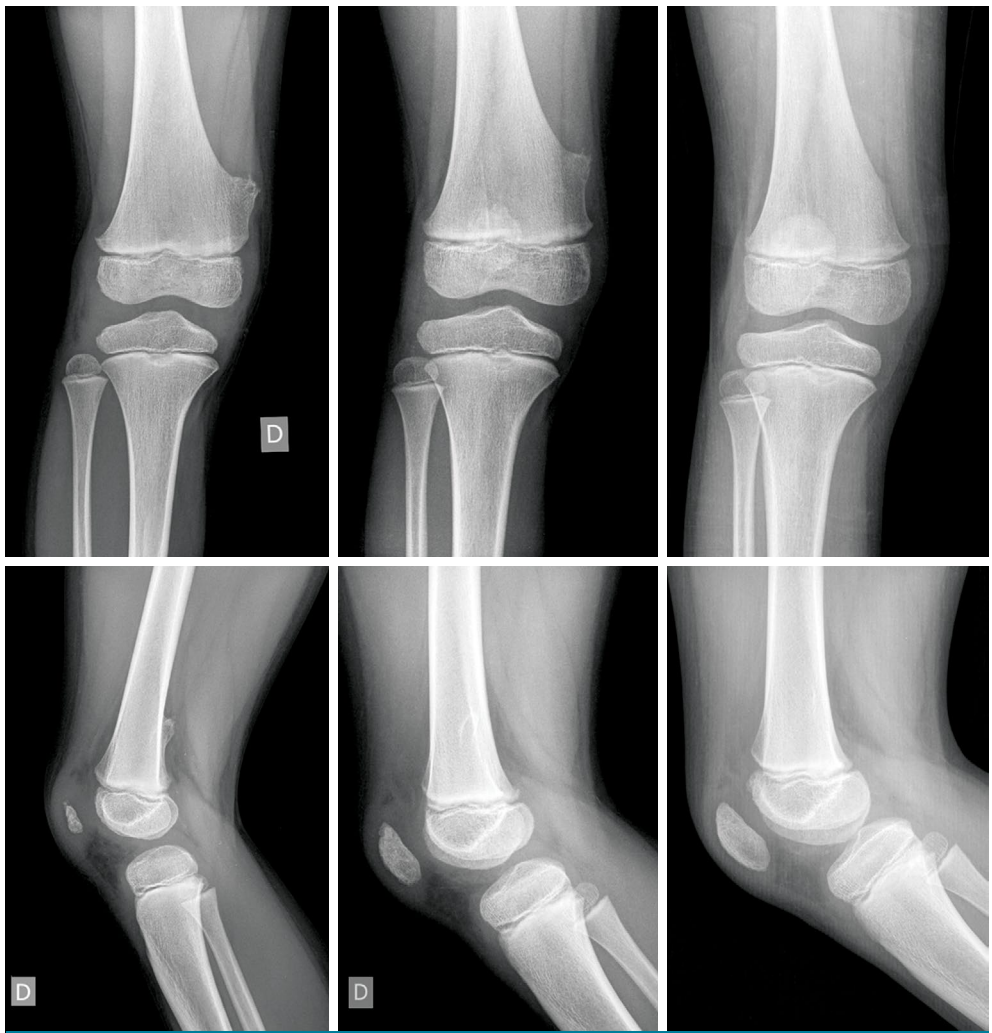

FIGURE 1. Anteroposterior and lateral X-rays of the right knee. A four-year-old boy with an osteochondroma in distal femur followed by X-ray annually, until the tumor vanished.

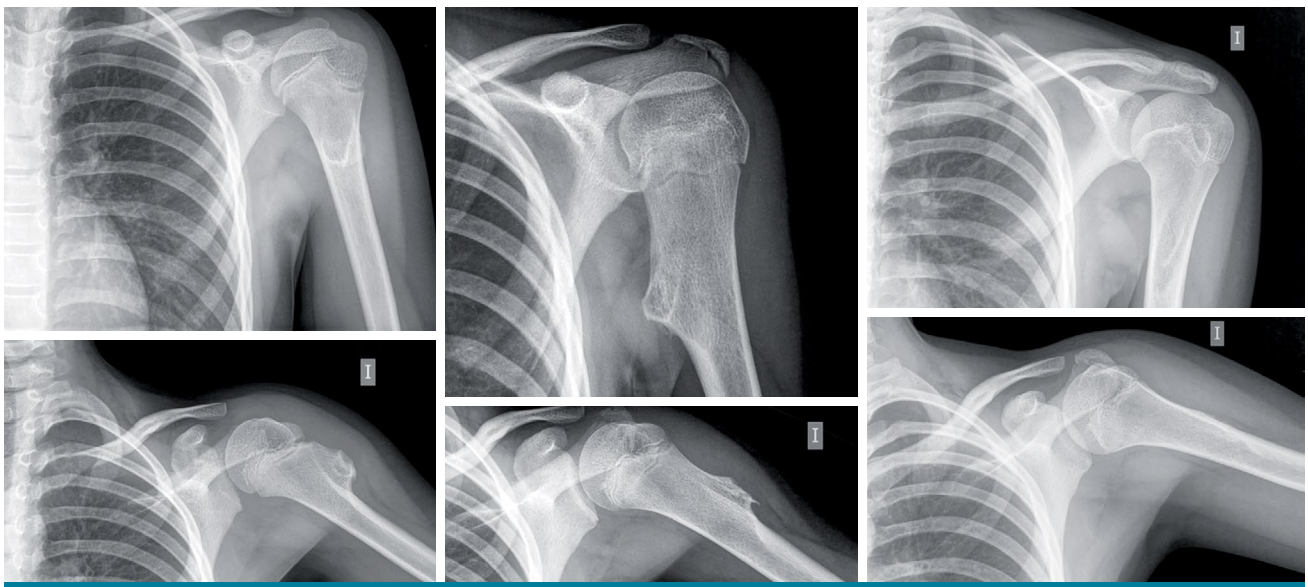

FIGURE 2. Anteroposterior radiographies of the left shoulder. A 10-year-old boy with an osteochondroma in proximal humerus followed by X-ray for three consecutive years.

upper arm. Radiographs showed a pedunculated osteochondroma on the proximal humerus. It measured 23.74 mm in diameter at the base and projected 17.25 mm away from the cortex (Figure 4). The patient did not report pain with regard to the osteochondroma, although the bony prominence was palpable. There was no neurovascular compromise. Annual follow-up showed clinical and radiographic spontaneous regression of the lesion within four years.

A written informed consent was obtained from the parents and/or legal guardians of the patient.

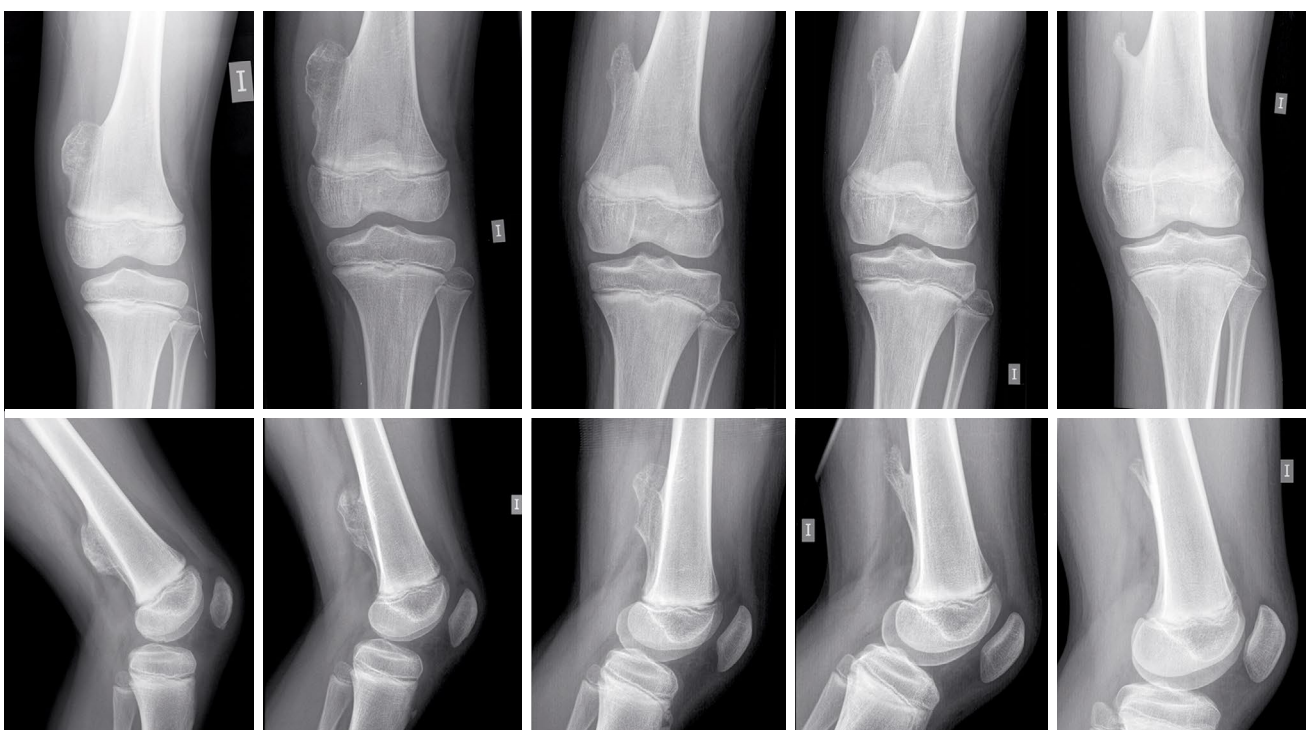

FIGURE 3. Anteroposterior and lateral X-rays of the left knee. A 10-year-old boy with an osteochondroma in distal femur followed by X-ray annually, until the age of 15 and the tumor almost vanished.

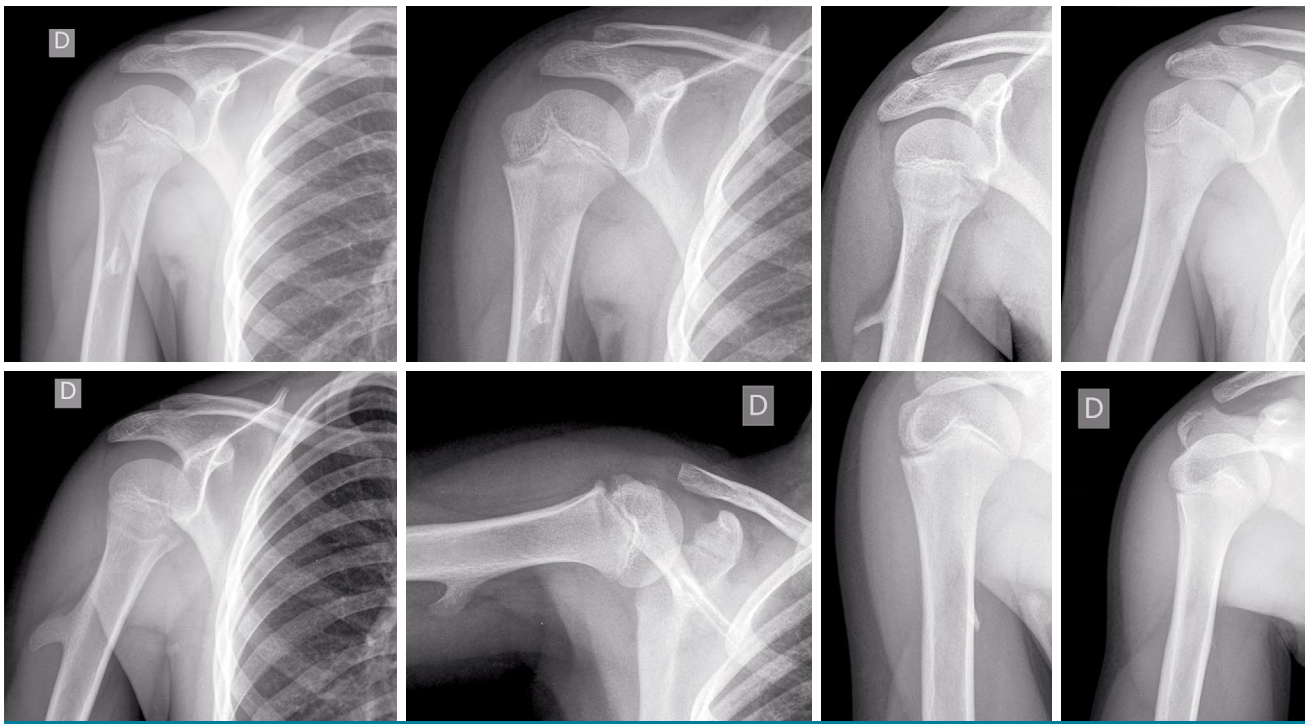

FIGURE 4. Anteroposterior radiographies of the right shoulder. An 11-year-old boy with an osteochondroma in proximal humerus followed by X-ray for four consecutive years.

DISCUSSION

According to Dahlin and Unni,[1] osteochondromas represent 40% of benign tumors and 10% of primitive bone tumors, being more frequent in men (ratio: 1.5/1).[2] Most of them are diagnosed in the second decade of life and usually stop growing after the closure of the bone physis, remaining commonly asymptomatic.[3] Symptomatic forms usually manifest before the age of 20 years, being pain and bony mass detection the most common symptoms.[3]

Osteochondromas are observed in bones with endochondral growth, at the level of the metaphyseal aspect of the fertile growth cartilages, in flat bones, and those of the axial skeleton. The discovery of a diaphyseal exostosis assumes that the lesion has migrated during growth.[3]

Sarcomatous degeneration of solitary osteochondromas is uncommon, and is around 1% of recognized exostoses.[1,4] These cases are usually accompanied by clinical exacerbation and radiographic changes.

Osteochondromas are benign tumors that usually have a slow and painless progressive growth. However, spontaneous regression is an exceptional event. Conservative treatment with clinical and radiographic annual follow-up may be a possible approach.[5]

In most cases, radiographs are sufficient to establish the diagnosis and follow-up.[5-17] Pathological diagnosis is not usually necessary. Nevertheless, Hoshi[18] used computed tomography (CT) and Minami[19] magnetic resonance imaging (MRI) to analyze the tumor. Deprez[20] employed radiographs and high-frequency ultrasound studies to measure the size and thickness of the cartilaginous cap of the lesion.

For efficient radiation protection, particularly in children, we consider that CT studies should be restricted to doubtful X-ray images, rapidly growing lesions, neurovascular compromise or possible malignant degeneration.[21,22] The study of the tumor cartilage cap or the surrounding soft tissues can be done combining X-ray with ultrasound, as it has a comparable value with CT.[23]

Magnetic resonance imaging is a valuable complementary test that can show specific characteristics of the cartilaginous cap of osteochondromas; however, the necessity of general anesthesia in children and its high cost make it a non-cost-effective tool for the diagnosis and

follow-up of osteochondromas in children. Most osteochondromas can be diagnosed and followed-up by simple X-ray. We recommend restricting ultrasounds, CT and MRI to doubtful X-ray images, rapidly growing lesions, neurovascular compromise or possible malignant degeneration.

According to the uncommon spontaneous regression of osteochondromas, we conducted a bibliographic research and rescued 16 case reports (Table I). Paling[6] referred that osteochondroma natural history depended upon the temporal relationship between its own growth and that of the host bone. This regression apparently resulted from a cessation of growth of the osteochondroma prior to skeletal maturation, with subsequent incorporation of the lesion into the enlarging bony metaphysis.[6] Copeland[7] was among the first to write about the

TABLE I
Cases of spontaneous regression of osteochondromas in literature

Case report	Year	No. patients	Location	Sex	Age at diagnosis (year)	Time until regression (year)	Trauma (Yes/No)
Paling[6]	1983	1	Distal femur	Male	9	6	No
Copeland et al.[7]	1985	2	Distal femur	Male	11	1.5	Yes
			Distal femur	Male	10	2	Yes
Castriota-Scanderbeg et al.[8]	1995	2	Distal radius	Male	12	1	Yes
			Proximal humerus	Female	5	3	No
Claikens et al.[9]	1998	1	Distal ulna	Male	7	0.5	No
Yamamoto et al.[10]	2001		Proximal phalanx	Male	3	7	No
Reston et al.[17]	2004	1	Distal femur	Male	15	4	No
Hoshi et al.[18]	2007	1	Proximal humerus	Male	7	1.25	No
Arkader et al.[11]	2007	1	Distal femur	Female	12	6	No
Minami et al.[19]	2009	1	Distal tibia	Female	6	2	No
Valdivielso-Ortiz et al.[12]	2010	1	Distal femur	Female	9	4	No
Mahmoodi et al.[13]	2010	1	Proximal tibia	Female	9	9	No
Deprez et al.[20]	2011	1	Distal femur	Female	11	4	No
Passanisse et al.[5]	2011	4	Humerus	Male	6	3	No
			Humerus	Male	7	5	No
			Humerus	Male	10	2.5	No
			Distal femur	Male	12	4	No
Hill et al.[14]	2014	1	Distal femur	Male	6	3	No
Heyworth and Rashid[15]	2018	1	Distal humerus	Male	1.25	0.25	Yes
Aiba et al.[16]	2018	8	Distal femur	Male	13	10	No
			Distal femur	Male	13	5	No
			Distal fibula	Female	13	3	No
			Distal femur	Male	14	4	No
			Distal tibia	Male	13	4	No
			Distal femur	Male	16	3	No
			Distal femur	Male	7	6	No
			Pelvis	Male	12	5	No
Our study	2021	4	Distal femur	Male	4	3	No
			Proximal humerus	Male	10	3	No
			Distal femur	Male	10	5	No
			Proximal humerus	Male	11	4	No

spontaneous resolution of osteochondromas. In 1985, he published two cases located in the distal femur. Copeland considered that the regression of the lesions might be attributed to cessation of the growth of the cartilaginous cap, followed by active resorption. He also related it to the appearance of a fracture line at the base of the exostosis that may have compromised the growth of the cartilaginous cap or altered its vascular supply. The fracture may have also stimulated growth in the periosteal tissue which functionally, then, resembled a normal periosteum, capable of mediating increased remodeling. Castriota-Scanderberg et al.[8] agreed with the Copeland's theory of bone resorption after exostosis fracture, since he presented a case with a similar pathogenesis. They suggested that, under the stimulus of trauma, the cells with osteogenic potencies in the periosteum became actively engaged in the remodeling of bone. In total, four of the cases revised mentioned a trauma antecedent[7,8,15] that could have triggered the shrinkage of the tumor and reduce the time to its regression; however, due to the inaccuracy between the time of exostosis appearance and its diagnosis, the exact time of regression is uncertain. In relation to the regression modes described by Aiba,[16] our cases were included in the incorporation theory, since the osteochondromas became mature before growth plate closure and were incorporated into bone growing in a vertical axis.

According to our bibliographic revision, we found 14 cases of osteochondroma regression in distal femur (48.28%) and five in proximal humerus (17.24%). These locations are the most commonly documented and this may be due to the higher incidence of osteochondromas in these areas.[24] All the cases presented in our study were in those locations.

In the literature, the average age at the time of diagnosis is 9.61 (range, 1.25 to 16) years and 13.51 (range, 1.5 to 23) years at the time of resolution.[5-16,18-20] In our study, the average age at time of diagnosis was 8.75 (range, 4 to 11) years and 12.5 (range, 7 to 15) years at the time of resolution. In addition, the four cases we present follow a similar pattern: they are all males in the first or second decade of their lives who noticed an unpainful bony mass in their extremities (proximal humerus and distal femur). They were followed by clinical examination and X-ray annually, until osteochondroma vanished.

In conclusion, although the initial rapid growth of the osteochondroma makes surgery tempting, it may not be necessary at all. If local symptoms are not severe and there is no evidence of malignant degeneration, it is reasonable to delay intervention and follow a conservative treatment, taking into account that spontaneous regression of solitary osteochondromas may occur.

Declaration of conflicting interests

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