



Case report

Osteopoikilosis with bony deformities of the right toes in a female child: Case report

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ABSTRACT

Introduction: Osteopoikilosis (OPK) is an extremely rare benign condition with sclerosing bony dysplasia and multiple benign enostoses. OPK is usually asymptomatic and is typically an incidental finding on imaging studies for unrelated conditions.

Case presentation: We presented a case of OPK in a 7-year-old female with hallux valgus, shortening and deformity of second and third metatarsals in the right foot. These abnormalities were observed on clinical findings with X-ray imaging, and osteopoikilosis was confirmed by histopathology. The deformities were treated with surgical intervention, and the patient's condition was followed for 3 months until the patient walked and removed the gypsum.

Discussion: OP is a rare, benign disease that rarely causes bony deformities. It is diagnosed clinically and radiographically, so that the deformities are treated only surgically. Follow-up is necessary to assess the movement of the limb.

Conclusion: The distinctive thing that can be added to the medical literature is that it is possible for osteopoikilosis to cause bone deformities at an early age.

1. Introduction

Osteopoikilosis (also called osteopathia condensans disseminata, spotted bones, osteosclerosis disseminata, osteosclerosis familiaris disseminata, and osteosclerosis fragilis generalisata) is a sclerosing bone dysplasia consisting of focal deposits of dense lamellar bone in the spongiosa [1].

Albers-Schonberg was the first to describe this uncommon sclerosing bone dysplasia in 1915 [2].

Osteopoikilosis is a rare inherited condition of the bones, transmitted as an autosomal dominant trait characterized by numerous hyperostotic areas that tend to localize in periarticular osseous regions. It is usually asymptomatic and it is often diagnosed incidentally during X-rays made by other reasons. It can occur at any age in both sexes [3].

Typically affects the small tubular bones of hands and feet, metaphyseal region of long bones, and pelvic bones. The hands are involved in nearly all patients, followed by the feet, pelvis, and long

bones [4].

According to the medical literature, Osteopoikilosis is not treated unless it is accompanied by deformities.

This case report has been reported in line with the SCARE criteria [8].

2. Case presentation

A 7-year-old female was admitted to our hospital with complaint the presence of right foot deformity. In clinical examination, the presence of hallux valgus, overlap of the second and third toes, deformity and shortening of the metatarsals were observed (Fig. 1).

The study of right foot X-ray revealed abnormal bone tissue structure (Fig. 2) and the findings of the clinical examination were confirmed. There is no family, medical, surgical or allergic history. The relative laboratory tests such as alkaline phosphatase, complete blood count (CBC) and Ca were within the normal ranges.

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Fig. 1. Hallux valgus, overlap of the second and third toes, and deformity and metatarsals of the metatarsals.



Fig. 2. The study of feet X-ray revealed abnormal bone tissue structures.

We did a bone scan of the whole body that showed irregular density structures with defined edges. They are located inside the epiphysis, do not destroy the bony cortex and do not exceed the growth plate, while the articular distances remain within the normal. They are located in the shoulder, hip and knee joints on both sides with the bones of the right and left ankle.

Then, we did an excisional biopsy of the left ankle with a size of (1.5 × 1 × 0.3) cm that showed segments of mature bony trabeculae with small fragments of mature cartilage tissue, also, malignancy was denied (Fig. 3).

According to the radiological and medical findings we confirmed the presence of osteopoikilosis. The decision about surgery was taken, three surgeons, an anaesthesiologist and a nurse participated in the surgery and they had difficulty using an adult technique for a child with a

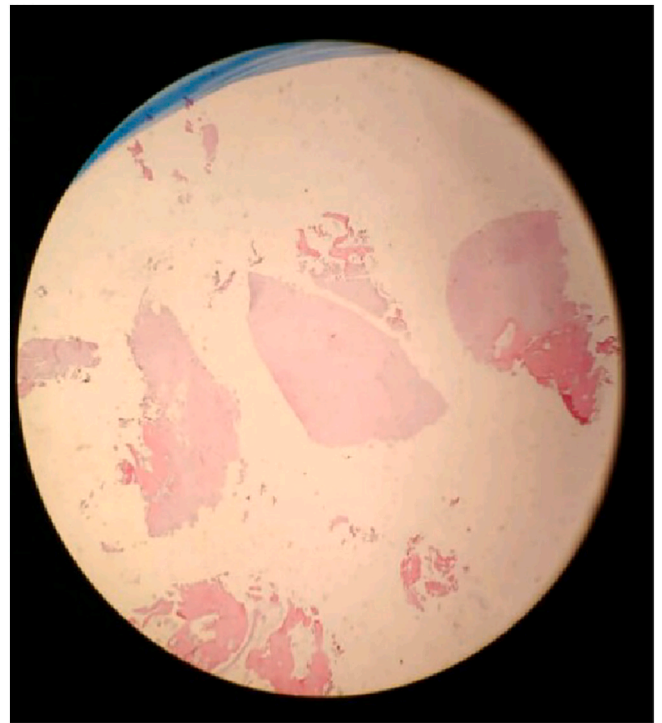


Fig. 3. Segments of mature bony trabeculae with small fragments of mature cartilage tissue.

functional metaphysis. The operation was performed under general anaesthesia with halothane and it took about 60 min, after the operation, paracetamol, ceftriaxone and metronidazole were used within the clinical dosage range to reduce the risk of pain and infection. The operation did not require drainage and no purulent oozing occurred. After the operation, we put a splint on the child's foot and discharged her after three days, then, the splint was removed after 6 weeks.

The child's condition was followed up for three months until the child was able to walk. No complications occurred and the results were acceptable.

3. Discussion

Osteopoikilosis (OpK) is a condition in which many small-roundish spots are found in the bones, particularly near the joints with sclerosing bony dysplasia and multiple benign enostoses. Osteopoikilosis may be inherited by autosomal dominant inheritance as part of Buschke ollendroff syndrome or sporadic (many occur randomly with no other features). It may be associated with synovial osteochondromatosis and melorheostosis.

The (OPK) incidence rate is estimated at 1/50,000 [5] and the incidence of men to women is 3/2 [6]. Benli et al. noticed that the most common sites of appearances for this condition were phalanges 100% and phalanges of the foot form 87,2% [7].

It is asymptomatic but rarely 15–20% patients feel slightly juxta articular pain and many occur joint effusion. In addition, it is detected by chance during imaging for another reason, as well as the bony lesions do not develop and are less seen in younger, thus it can be missed early in life. But in our case, It occurs at a younger age and leads to abnormalities. OPK is diagnosed by performing an X-ray and excisional biopsy especially as there are clinically detected abnormalities. As a result, to distinguish from bone metastases or other metastases, the diagnosis is confirmed by excisional biopsy. OPK in medical literature is not treated unless it is associated with deformities and the treatment for this condition is surgical. When this condition occurs in child, a special technique must be used because the child has an effective epiphyseal line

and when treating such a case, there must be follow-up until the child walks and it must be confirmed that his walking is normal.

4. Conclusion

Osteopoikilosis (OPK) is an extremely rare benign condition with autosomal dominant inheritance or sporadic with sclerosing bony dysplasia, it is asymptomatic and detected by chance during unrelated other reason.

This should be kept in mind that (OPK) may be cause abnormalities and it should be treated with orthopedic surgery.

Declaration of competing interest

All authors declare no conflict of interest.

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Consent

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journal on request.

Author contribution

Sarya Swed: contributed in study concept and design, data collection, and writing the paper.

Alaa Baria: contributed in writing the paper.

Mahmoud Alswij: contributed in writing the paper.

Hiba Haj saleh: contributed in writing the paper.

Jamal Katnaji: contributed in reviewing the paper.

Waleed Abd: contributed in reviewing the paper.

Research registration

Not applicable.

Guarantor

Sarya Swed.

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