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Editorial

Odd and Offbeat-Bizarre Parosteal Osteochondromatous Proliferation

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Received: July 02, 2025; **Published:** August 07, 2025

Bizarre parosteal osteochondromatous proliferation emerges as a benign neoplasm frequently confined to bony surfaces of small bones of hands and feet. Initially scripted by Nora in 1983, neoplasm demonstrates an exophytic pattern of bony outgrowths superimposed with an intact bone cortex. However, additional terminology of Nora lesion is not recommended [1].

Tumefaction is comprised of an admixture of bland, spindle shaped cells configuring bundles of disorganized cartilage commingled with benign bony tissue and a characteristic, basophilic matrix denominated as 'blue bone' Tumefaction emerges as a cellular lesion comprising of disorganized bundles of spindle shaped cells commingled with atypical chondrocytes and bone.

The lesion commonly arises within young individuals or adult subjects, especially within third decade to fourth decade although no age of disease emergence is exempt. A specific gender predilection is absent.

Bizarre parosteal osteochondromatous proliferation typically implicates small bones of hands and feet. Hands are frequently involved followed in frequency by feet and long bones. Exceptionally, craniofacial bones may display the lesion [2,3].

Tumefaction depicts repetitive chromosomal anomalies as genetic translocation t(1;17)(q32-42;q21-23) and inv(7)(q21.1-22q31.3-32). Chromosomal anomalies appear concurrent with COL1A2::LINC-PINT and COL1A1::MIR29B2CHG genetic fusions. Aforesaid molecular features induce disruption of type I collagen during tumorigenesis.

Neoplasm depicts reoccurring cytogenetic anomalies as chromosomal translocation t(1;17)(q32;q21), inv(7) and inv(6). Notwithstanding, aforesaid genomic alterations appear diverse from chromosomal modifications encountered within subungual exostoses [2,3].

Benign parosteal osteochondromatous proliferation is posited to emerge from a disease spectrum comprised of lesions as florid reactive periostitis or turret exostosis. Alternatively, neoplasm is postulated to occur on account of repetitive cytogenetic anomalies. Additionally, lesion may possibly arise due to traumatic aetiology [2,3].

Clinical symptoms are characteristic and a gradually progressive, firm, painless tumour nodule of few months to several years duration is encountered. Generally, tumour nodule appears as a miniature lesion of one centimetre to 3 centimetre magnitude. Few instances demonstrate brisk tumour progression, indicative of possible malignant transformation. Localized symptoms may appear due to mass effect. Few lesions may be accompanied by preceding history of trauma [3,4].

Cytological smears are cellular and composed of proliferating spindle shaped cells. Focal calcification is encountered. Cellular and nuclear atypia is insignificant. Grossly, an exophytic bony lesion is observed wherein the bony matrix is superimposed with cartilage.

Tumour magnitude preponderantly varies from one centimetre to 3 centimetres [3,4].

Upon microscopy, tumefaction is comprised of quantifiably variable admixture of cartilage, bone and fibrous tissue configuring disorganized bundles.

Fascicles of disorganized cartilage configure an extraneous layer which is constituted of enlarged, atypical chondrocytes imbued with enlarged nuclei. Tumour cells may be bi-nucleated. The cellular tumefaction depicts transition into irregular interface of bony trabeculae through endochondral ossification [4,5].

Constituent bone configures benign appearing trabeculae which appear contiguous with subjacent cortex. Characteristically, tissue between cartilage and bone is comprised of basophilic stroma and is thereby denominated as 'blue bone'.

Constituent cellular fibrous tissue is loosely disseminated and comprised of spindle shaped to stellate cells intermixed between bony trabeculae. Cytological atypia is minimal. Atypical mitotic figures appear absent [4,5].

TNM staging of malignant bone tumours as per American Joint Committee on Cancer 8th edition [4,5].

Primary tumour

Appendicular skeleton, trunk, skull, facial bones:

- TX: Primary tumour cannot be assessed.
- T0: No evidence of primary tumour.
- T1: Tumour is ≤ 8 centimetre magnitude.
- T2: Tumour is > 8 centimetre magnitude.
- T3: Primary tumour site exhibits multi-centric neoplasms.

Spine:

- TX: Primary tumour cannot be assessed.
- T0: No evidence of primary tumour.
- T1: Tumour is confined to single vertebral segment or singular focus of vertebrae or two adjacent vertebral foci.
- T2: Tumour is confined to three adjacent vertebral segments.
- T3: Tumour is confined to ≥ 4 adjacent vertebral segments or non adjacent vertebral segments.
- T4: Tumour invades spinal canal or great vessels of vertebral column:
- T4a: Tumour extends into spinal canal.
- T4b: Tumour invades great vessels of vertebral column or impedes vascular outflow.

Pelvis

- TX: Primary tumour cannot be assessed.
- T0: No evidence of primary tumour or extra-osseous extension.

- T1: Tumour manifests singular focus within pelvis.
- T1a: Tumour is ≤8 centimetre magnitude.
- T1b: Tumour is > 8 centimetre magnitude.
- T2: Tumour is confined to singular pelvic focus with extra-osseous extension or two pelvic foci with absent extra-osseous extension.
- T2a: Tumour is ≤8 centimetre magnitude.
- T2b: Tumour is >8 centimetre magnitude.
- T3: Tumour is confined to two pelvic foci with extra-osseous extension.
- T3a: Tumour is ≤8 centimetre magnitude.
- T3b: Tumour is > 8 centimetre magnitude.
- T4: Tumour is confined to three pelvic foci or extends beyond sacroiliac joint.
- T4a: Tumour invades sacroiliac joint and incriminates sacral neuro-foramina.
- T4b: Tumour encases regional vasculature or impedes vascular outflow.

Regional lymph nodes

- NX: Regional lymph nodes cannot be assessed.
- N0: Regional lymph node metastasis absent.
- N1: Regional lymph node metastasis present which is exceptional in a primary bone sarcoma.

Distant metastasis

- M0: Distant metastasis absent.
- M1: Distant metastasis present:
- M1a: Metastasis into pulmonary parenchyma.
- M1b: Metastasis into various bones or viscera.

Bizarre parosteal osteochondromatous proliferation lacks distinctive immunoreactivity to diverse biomarkers. CD68 may be adopted to highlight disseminated osteoclasts. Cartilaginous zones appear variably immune reactive to Safranin O.

Tumour cells appear immune non reactive to MDM2 [5,6].

Bizarre parosteal osteochondromatosis proliferation requires segregation from neoplasms as florid reactive periostitis, myositis ossificans, fibro-osseous pseudotumour of digits, Turret exostosis, subungual exostosis, periosteal chondroma, osteochondroma, parosteal osteosarcoma, periosteal chondrosarcoma and periosteal osteosarcoma [5,6].

Ultrasonography depicts a calcified soft tissue tumefaction which lacks extension into subjacent cortex [6,7].

Upon imaging, an exophytic bony tumour demonstrates an intact bony cortex. Neoplasm is composed of three distinct components designated as spindle shaped cells, bone and atypical cartilage [6,7].

Upon radiography, a preliminary stage of periosteal soft tissue swelling is followed by progressive mineralization of bone whereas the delayed stage enunciates an ossified tumefaction confined to bone surface.

Computerized tomography (CT) delineates an ossified, contiguous neoplasm confined to bone surface delineating non continuity with subjacent cortex. However, a distinctive discontinuity of bony cortex or specific tumour extension into the bone marrow appears absent. Infrequently, tumefaction may display continuity with medullary cavity [6,7].

Magnetic resonance imaging (MRI) depicts a hyper-intense peripheral signal intensity on account of cartilaginous component.

T1 weighted imaging depicts minimal to intermediate signal intensity.

T2 weighted imaging displays intermediate to enhanced signal intensity.

Images may appear enhanced upon administration of contrast medium.

Asymptomatic bizarre parosteal osteochondromatous proliferation may be appropriately subjected to conservative management. Neoplasm may be managed with simple surgical extermination [6,7].

Localized tumour reoccurrence may be appropriately treated with repetitive surgical eradication or surgical resection with removal of a broad perimeter of uninvolved tissue [6,7].

Neoplasm is accompanied by benign clinical course although tumour reoccurrence appears frequently in ~ 55% lesions. Multiple recurrences may be encountered [6,7].

Tumour reappearance is non destructive and may be suitably managed by repetitive surgical extermination. Neoplasm appears to lack a distinctive possibility of distant metastasis [6,7].

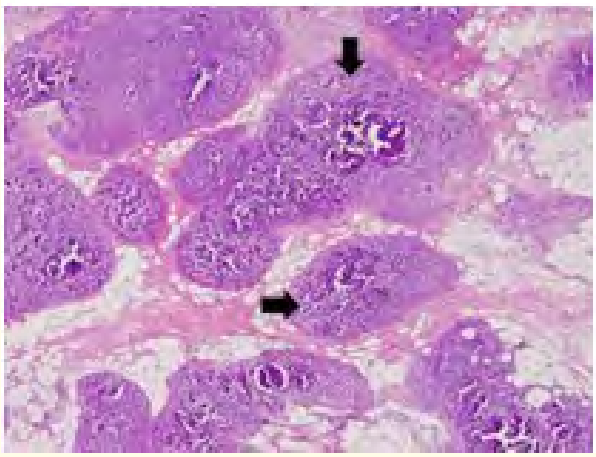

Figure 1: *Bizarre parosteal osteochondromatous lesion delineating an admixture of bone, cartilage and fibrous tissue. Bundles of disorganized cartilage is superimposed upon bony cortex and foci of blue bone. Intervening fibrous tissue is loose [8].*

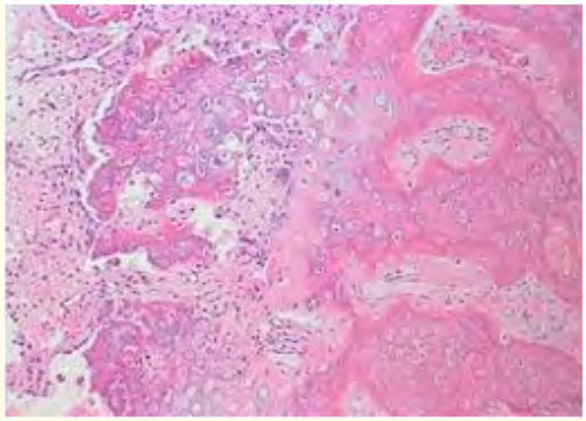

Figure 2: *Bizarre parosteal osteochondromatous lesion delineating commingled bone, cartilage and fibrous tissue. Disorganized chondrocytes appear superimposed upon bony trabeculae and blue bone and are encompassed within a loose fibrotic stroma [9].*

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8. Image 1 Courtesy: Science direct.

9. Image 2 Courtesy: Libre pathology.

Volume 17 Issue 9 September 2025