Impact of Structured Reporting of Skeletal Survey in Skeletal Dysplasia: A Single Institution Experience

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Abstract

Background Structured reporting has the advantages of reducing ambiguity in written radiology reports with greater uniformity and comparability of reports amongst different institutes. It has multiple facets: structured format, structured content, and standardized language. While structured reporting initiative has been used in various radiology subspecialties such as oncology, cardiothoracic, abdominal and interventional radiology; skeletal dysplasia is a domain that remains largely untouched by this concept.

Purpose   To evaluate the impact of structured reporting in skeletal dysplasia.

Methods and Materials   This was an ethically approved pragmatic clinical trial. A defined number (75) of clinically diagnosed and/or genetically confirmed skeletal dysplasia radiographs were evaluated by two radiologists (reader A and reader B) with 5-and 7-years' experience in general radiology, respectively. A pre-defined structured reporting format for skeletal dysplasia was used as an interventional tool. Both the readers interpreted the radiographs before and after the training session. In addition to diagnosis, diagnostic confidence was noted using a semiquantitative scale. Improvement in diagnostic accuracy and diagnostic confidence after training were assessed. McNemar's test was used to assess the statistical significance of difference in proportion of correct diagnoses in pre- and post-education phases. An interrater reliability analysis using the Kappa statistic was performed to determine interobserver agreement between readers both in pre- and post-education phases.

Results   In the post-education phase, the proportion of accurate diagnosis improved from 48% (36/75) to 64% (48/75) for reader A, and from 44% (33/75) to 60% (45/75) for reader B as compared with the pre-education phase. Amongst the cases with a correct radiologic diagnosis, an increase in diagnostic confidence was noted in 18 cases for reader A, and 15 cases for reader B. In none of the cases, there was a reduction in diagnostic confidence after training. A McNemar's test determined that there was a statistically significant difference in the proportion of correct diagnoses in pre- and post-education phases, $p < 0.001$. The interobserver agreement between the readers

Keywords
► skeletal dysplasia
► structured reporting
► skeletal survey

was found to increase from Kappa $= 0.33$ ($p = 0.004$) using non-structured reporting in pre-education phase to Kappa $= 0.46$ ($p < 0.001$) using structured reporting in the post-education phase.

Conclusion   A structured reporting of skeletal survey can improve accuracy and confidence in diagnosing skeletal dysplasia.

Introduction

Radiology reports are an important health record, and there is a lot of ongoing discussion regarding the topic of structured reporting.[1,2] As an endeavor toward promoting uniformity in reporting, structured reporting is an important step. Structured reporting has the advantages of reducing ambiguity in written radiology reports with greater uniformity and comparability of reports amongst different institutes. Structured reporting has multiple facets: structured format, structured content, and standardized language.[3] Format structuring can be done by dividing it into different well-defined sub-parts (such as clinical details, imaging parameters, imaging findings, and radiological diagnosis). The next tier of structuring can be done in the content (for example, description of each bone in a skeletal survey, or description of each structure in a chest scan). Finally, the way of describing the findings should be standardized. Standardized terminologies for various disease entities can be found in various lexicons (www.radlex.org), or various society guidelines.

While structured reporting initiative has been used in various radiology subspecialties such as oncology, cardiothoracic, abdominal and interventional radiology, skeletal dysplasia is a domain that remains largely untouched by this concept.

Purpose of Study

To evaluate the impact of structured reporting in skeletal dysplasia.

Methodology

We undertook this study after obtaining ethical clearance from the institutional ethics committee. This was a pragmatic clinical trial. A defined number ($n = 75$) of skeletal surveys were given for reporting to two radiologists (reader A and reader B) with 5- and 7-years' experience in general radiology, respectively. The patients whose complete skeletal survey images were available in the departmental archives and where a clinical/clinico-radiological/genetically proven diagnosis of skeletal dysplasia was made were enrolled for reporting. Patients with incomplete imaging data or inconclusive working diagnosis were excluded from the study.
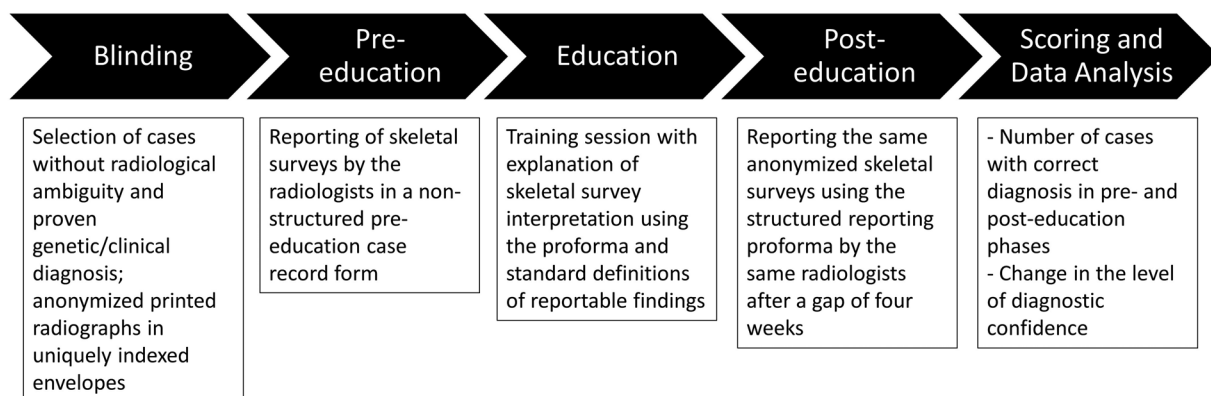
A pre-defined structured reporting format for skeletal dysplasia was used as an interventional tool.

Methodology and workflow is shown in ►**Fig. 1**.

At the beginning, a reading set of radiographs was prepared using the skeletal surveys accessed from the electronic or physical archives of the department. Clinical records of the patients were recovered from the hospital information system. A final confirmed diagnosis was noted against all skeletal surveys.

Blinding

The department has a picture archiving & communication system (PACS), with an online reporting portal, where patient reports are searchable with a unique hospital identification number (UHID). Hence, printed radiographs of patients who fulfilled the diagnostic criteria were provided in separate uniquely indexed envelopes. This indexing was different from the UHID of the patient, thus making it impossible to search for previously generated reports of the patient. This entire process was performed by investigator Y (not involved in reporting). Investigator Y also maintained a master key enabling conversion between the unique indexing and UHID of the patient. The only information not blinded were the age and gender of the patients.

Fig. 1 Study methodology and workflow.

Blinding	Pre-education	Education	Post-education	Scoring and Data Analysis
Selection of cases without radiological ambiguity and proven genetic/clinical diagnosis; anonymized printed radiographs in uniquely indexed envelopes	Reporting of skeletal surveys by the radiologists in a non-structured pre-education case record form	Training session with explanation of skeletal survey interpretation using the proforma and standard definitions of reportable findings	Reporting the same anonymized skeletal surveys using the structured reporting proforma by the same radiologists after a gap of four weeks	- Number of cases with correct diagnosis in pre- and post-education phases - Change in the level of diagnostic confidence

Pre-Education Phase

Reader A (resident with 5 years' experience) and reader B (pediatric radiology fellow with 7 years' radiology experience) were instructed to go through any educational resource, which they find valuable before beginning the reporting. A time of 1 week was given for the same. Then, the two readers were separately given the uniquely indexed, blinded cases for review along with patient age and gender. Our study did not involve skeletal dysplasia mimics, and hence all cases were those of skeletal dysplasias. The same was conveyed to both readers A and B. They then reported the cases in a routine prose format ("pre-education case record form") digitally over a 1-week period. In this period, they were instructed not to refer to any educational materials or discuss their findings with other radiologists (including between themselves). None of them were given the proposed structured reporting format before the education phase.

Diagnostic confidence in reporting the cases was recorded using the following semiquantitative scale:

0: unable to diagnose
1: can narrow down to three to four differential diagnoses with confidence or can diagnose a group of disorders with confidence (for example, metaphyseal dysplasia, spondylometaphyseal dysplasia, etc.).
2: can narrow down to two differential diagnoses with confidence.
3: can give a single diagnosis with confidence.

The filled pre-education case record forms were submitted to investigator Y.

Education Phase

Investigator X (M.J., 12 years of pediatric radiology experience) introduced both the readers to the structured reporting format (►**Appendix A**). Operational standard definitions of reportable findings were also discussed. A training dataset consisting of 30 other separate skeletal surveys were used, consisting of both normal and abnormal skeletal surveys.

This phase was conducted as an in-person meeting over 3 hours. Specific cases used in pre-education phase were not discussed in this meeting.

Post-Education Phase

After a gap of 4 weeks, readers A and B filled the structured reporting format ("post-education case record form") digitally for the same set of skeletal surveys. In this period, they were not allowed access to any other educational materials or discussion of their findings with other radiologists (including between themselves). This phase was completed over a few staggered sessions over a week. After filling the same, the post-education case record forms were submitted to investigator Y (P.G.).

Scoring and Analysis Phase

At the end of the post-education phase, investigator Y had four sets of record forms (two sets of pre-education and two sets of post-education case record forms). These sets were randomly labeled as sets 1 to 4. Thereafter, investigator X scored the response sheets, after converting the unique indexing numbers to their original UHIDs using the master key available with investigator Y. The reader's diagnosis was compared with the patient's final diagnosis and note was made whether correct diagnosis was given for a particular case or not (yes/no format). The confidence in scoring was also noted as described using the semiquantitative scale. After scoring of the datasets by investigator X, correct relabeling of the sets was done by investigator Y, and descriptive data analysis was done. McNemar's test was used to assess the statistical significance of difference in proportion of correct diagnoses in pre- and post-education phases. An interrater reliability analysis using the Kappa statistic was performed to determine interobserver agreement between investigators both in pre-education and post-education phases.

Results

Out of the total of 75 cases of skeletal dysplasia used for reporting in this study, 42 were male and 33 were female. The median age of patients was 2.5 years (range, 1 month-15 years). The final diagnoses of the cases ($n = 75$) are shown in ►**Table 1**. In post-education phase, the proportion of accurate diagnosis improved from 48% (36/75) to 64% (48/75) for reader A, and from 44% (33/75) to 60% (45/75) for reader B as compared with pre-education phase (►**Fig. 2**). Amongst the cases with a correct radiologic diagnosis, an increase in diagnostic confidence was noted in 18 cases for reader A, and 15 cases for reader B. In none of the cases, there was a reduction of diagnostic confidence after training (►**Table 2**). A McNemar's test determined that there was a statistically significant difference in the proportion of correct diagnoses in pre- and post-education phases, $p < 0.001$. The interobserver agreement between the readers was found to increase from Kappa $= 0.33$ ($p = 0.004$) using nonstructured reporting in the pre-education phase to Kappa $= 0.46$ ($p < 0.001$) using structured reporting in the post-education phase.

Of note, entities showing closely overlapping radiological appearances (for example, Pseudoachondroplasia vs. Mucopolysaccharidoses) showed a better diagnostic accuracy and confidence with usage of standard terminology and structured reporting (►**Figs. 3** and **4**). Also, rare syndromes with unique radiographic appearances were better diagnosed in the post-education phase (►**Fig. 5**). Relatively common and widely described entities (such as achondroplasia, cleidocranial dysostosis, osteopetrosis) were diagnosed accurately and confidently by the readers in both the pre-education and post-education phases.
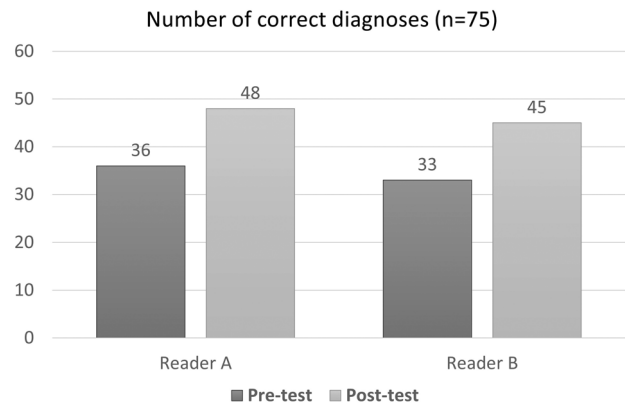
Discussion

Skeletal dysplasias are a heterogeneous group of disorders, for which multiple classification systems exist. There are myriads of manifestations and variants of each single disease entity, with many having various subtypes. Along with the correct diagnosis, reporting of the various specific manifestations is also important as they may guide further tailoring

Table 1 Final diagnoses of the skeletal surveys used in the study

Diagnosis	Number of cases
Achondroplasia	7
Hypochondroplasia	1
Pseudoachondroplasia	6
Progressive pseudo-arthropathy of childhood	6
Spondyloepiphyseal dysplasia congenita	8
Mucopolysaccharidoses	2
Mucolipidoses	2
Dyggve–Melchior–Clausen Syndrome	2
Jeune's asphyxiating thoracic dystrophy	2
Cleido-cranial dysostosis	2
Osteogenesis imperfecta	1
Ellis-van-Creveld syndrome	1
3M syndrome	2
Hereditary multiple exostoses	1
Multiple epiphyseal dysplasia	1
Congenital hypothyroidism	3
Fibrodysplasia ossificans progressiva	1
Spondylocostal dysostosis	1
Campomelic dysplasia	1
Spondyloepiphyseal dysplasia tarda	1
Enchondromatosis	1
Achondrogenesis	1
Spondylo-epi-metaphyseal dysplasia–Strudwick type	1
Spondylo-epi-metaphyseal dysplasia–Sutcliffe type	1
Spondylo-epi-metaphyseal dysplasia–Kozlowski type	1
Osteopetrosis	3
Osteopoikilosis	1
Osteopathia striata	1
Chondro-dysplasia punctata	3
Larsen syndrome	2
Diastrophic dysplasia	1
Melnick–Needles syndrome	1
Metaphyseal chondrodysplasia	4
Fibrous dysplasia	2
Pyknodysostosis	1
Total (n)	75

Number of correct diagnoses (n=75)

Fig. 2 Number of correct diagnoses made by Reader A and Reader B in pre-education and post-education phases respectively.

Table 2 Data showing the observer performance in post-education phase of the study compared with the pre-education phase

	Reader A (n = 75)	Reader B (n = 75)
Number of cases showing improvement in correct diagnosis	12	12
Number of cases showing improvement in diagnostic confidence (amongst cases with correct diagnosis)	18	15
Number of cases showing no change in diagnosis	21	25
Number of cases showing decrease in diagnostic confidence (amongst cases with correct diagnosis)	0	0

of patient-specific treatment. The most comprehensive and widely accepted classification for skeletal dysplasia has been given by the International Skeletal Dysplasia Society (2019).[4] This classification is based on genetics and is of limited use to the reporting radiologist. Nevertheless, making a radiologic diagnosis closest to the subset of disease often remains the goal.

Imaging in skeletal dysplasia largely depends on a skeletal survey. Skeletal survey contains a group of standardized radiographs of the axial and appendicular skeleton. Much unlike other types of imaging modalities (e.g., ultrasound, CT, and MRI), radiographs in a skeletal survey deliver mostly uniform set of data. Hence, implementation of a standardized reporting is less dependent on the imaging acquisition variables.

Mostly, the radiology report in a skeletal survey is written in a 'prose' manner; and it is possible to miss out one or more subtle imaging findings. While the major diagnostic features may not be omitted, the minor/less commonly described findings can go unnoticed owing to a 'satisfaction of search.' Reporting of such minor findings can actually make a significant difference as these may be the first pointer toward the detection of a new phenotypic/genotypic variant of a described disease entity. Moreover, detailed reporting of all
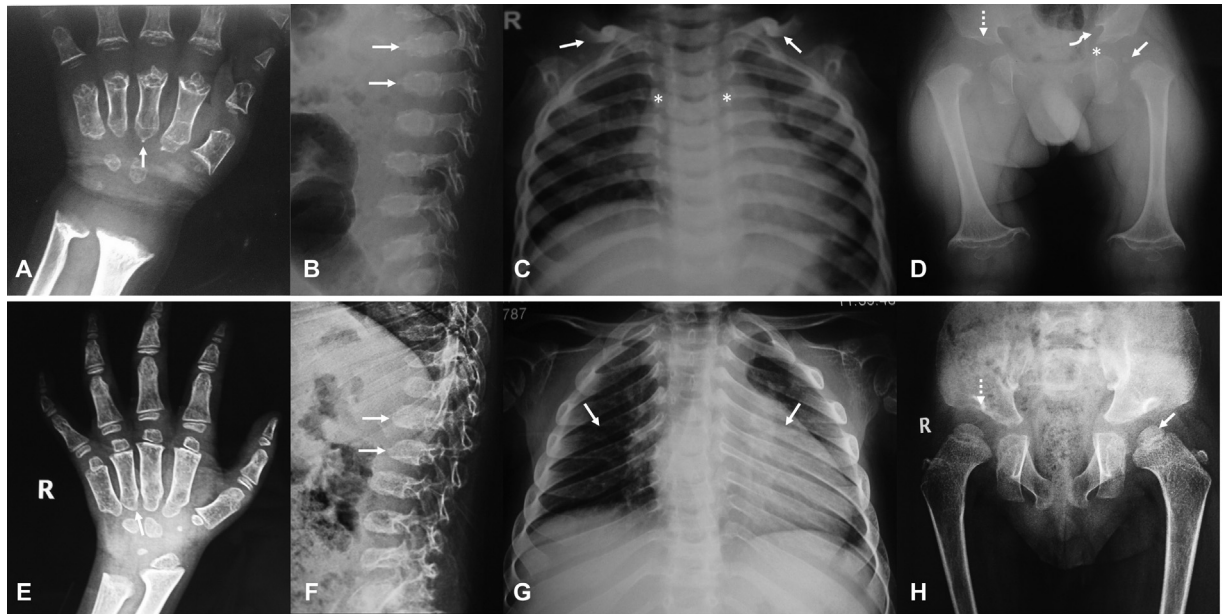
Fig. 3 Skeletal surveys of two patients diagnosed with pseudoachondroplasia (Patient A, a-d) and Hurler's syndrome (Patient B, e-h). Both patients show proximally pointed metacarpals (arrows in a and e) and central beaking of vertebrae (arrows in b and f), which lead to difficulty in reaching a conclusive diagnosis. However, thoracic and pelvic radiograph findings help in the correct diagnosis. Widened costovertebral junctions (asterisks in c), foreshortened clavicles (arrows in c), wide triradiate cartilage (asterisk in d), narrow sciatic notch (curved arrow in d), irregular horizontal acetabulum (dotted arrow in d) and very delayed appearance of femoral heads (arrow in d) point toward pseudoachondroplasia. Paddle-shaped ribs (arrows in g), sloping acetabulum (dotted arrow in h), and relatively preserved femoral heads (arrow in h) point toward Hurler's syndrome.

findings reduces probability of misdiagnosis as another pathology having overlapping clinical and imaging findings.

The presence of an enormous number and variants of dysplasias makes it humanly impossible to remember the details of each required to make a diagnosis. Hence, geneticists mostly refer to specific medical databases (such as London Medical Database). The search in such databases is dependent on selection of the right descriptive word. For example, search using 'platyspondyly' (reduced vertebral height) and 'anisospondyly' (variation in sizes of vertebrae) will not necessarily yield the same result. Herein lies the utility of using standardized terminology in skeletal dyspla-

Fig. 4 Sample of the structured reporting format for the same patients (Patients **A** and **B**) as in Fig. 2. Reporting of each pelvic and thoracic structure in a format using standardized terminology helps in reaching the correct diagnosis.

sia reporting. It is much more convenient to search a database using the standard language. For example, 'acromelia' will narrow down the search more efficiently to only those diseases having acromelic shortening, compared with 'shortening of limb length' or 'distal limb shortening.' Unfortunately, in pediatric radiology practice, very often the trainees are not exposed to an exhaustive structured reporting of a suspected skeletal dysplasia.

Another unique aspect of structured reporting in dysplasia is that it enables the radiologists to reach a reasonable set of differential diagnoses, using a step-wise or segment-wise approach. Interpretation of a skeletal survey is tough and needs experience. While a seasoned pediatric radiologist may not find it difficult to reach a diagnosis or a limited differential diagnosis, it is the trainees in pediatric radiology who need to master the art by adopting a more systematic way.[5]
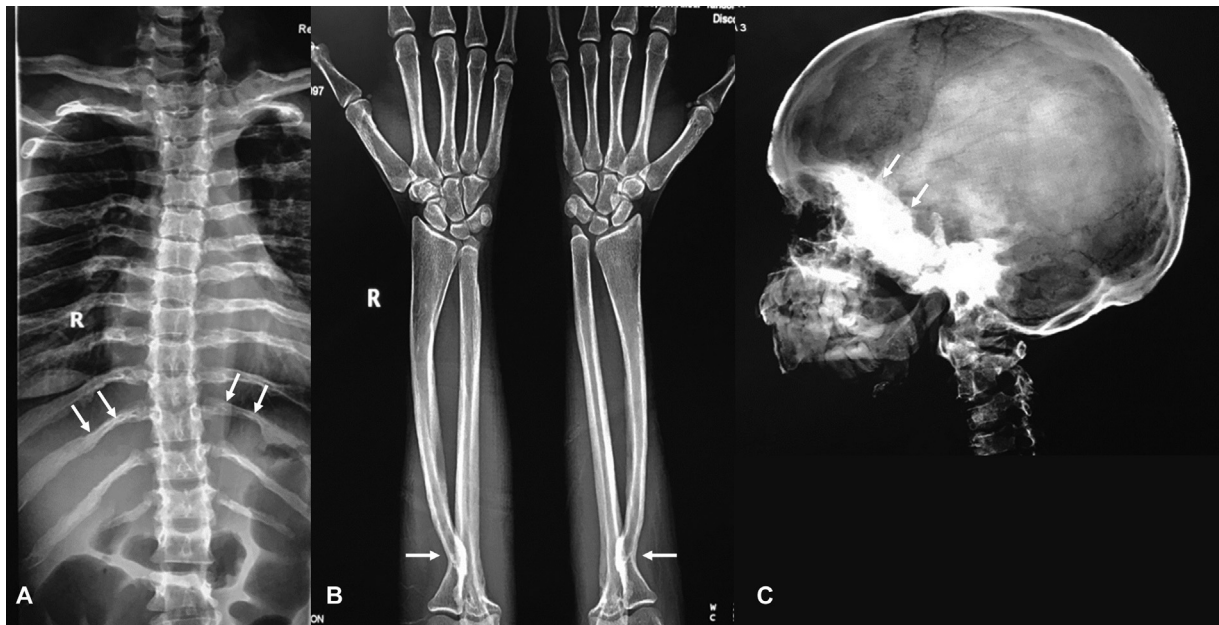
The Radiological Society of North America has a long list of accepted structured reporting templates, and there is one proposed for skeletal dysplasia too (available at www.radreport.org). Nevertheless, it is the beginning point in structured reporting of skeletal dysplasia, and a much more detailed format having a structured content is the need of the hour. An example of this can be presented as follows:

The description–'A child with short stature, hip and back pain, shows normal bone density, uniform platyspondyly, and normal bone age (taken at the carpal bones)' will probably not lead the radiologist (or the geneticist who is reading the report) to a definitive diagnosis. Similarly, a database search using these imaging findings would yield a long list of diseases. However, if a structured reporting format mandates the

Fig. 5 Unique radiographic findings in a rare skeletal dysplasia: Melnick–Needles syndrome. Ribbon shaped irregular ribs (arrows in a), diaphyseal bowing of radii (arrows in b), and skull base sclerosis (arrows in c) are characteristic findings. The diagnostic accuracy for such unique findings improved with the use of structured reporting format after education phase.

inclusion of femoral head size, and appearance of pubic bones, making a diagnosis of spondyloepiphyseal dysplasia congenita (SEDC) would become easier. A small femoral head size compared with the chronological age, and absence/delay in pubic bone ossification would serve as important key pointers in making the diagnosis in this case.

The disadvantages of structured reporting are also widely debated upon. It reduces the report to a set of values and descriptive terms, and leaves much less scope for the radiologists to exert their diagnostic skill. However, this argument is probably not apt in cases of skeletal dysplasia, where the diagnosis is most often a mathematical mix and match of radiological and clinical features. Such a match can only be achieved if we have noted all the findings. Hence, the need for an elaborate structured reporting and proper terminology in reporting a skeletal survey.

Our study had a few limitations. Only a small sample size could be utilized in the study owing to the rarity of skeletal dysplasias and difficulty in establishing a single confirmed diagnosis in these cases. Our skeletal surveys did not include any normal studies and other mimics of dysplasias (such as rickets and non-accidental injury), which could have led to overreporting of findings owing to bias. Also, this is unlike the routine clinical scenario wherein ruling out other metabolic diseases and mimickers of dysplasias is a pre-requisite for correct interpretation of findings.

To conclude, structured radiological reporting format using standardized terminology for various findings in skeletal dysplasia may help in narrowing down of differential diagnoses more efficiently and may provide a step-wise approach for a radiology trainee to approach this vast gamut of diseases.

Funding
None.

Conflict of Interest
None declared.

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