# Case report: Segmental odontomaxillary dysplasia

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## BACKGROUND

Segmental odontomaxillary dysplasia (SOD) is a rare, nonhereditary developmental disorder usually affecting the posterior maxilla. Clinical presentation consists of unilateral, painless expansion of the alveolar process with hypoplastic or missing premolar teeth in the affected region.

## CASE REPORT

### Clinical findings

A 7-year old female was referred by a general dental practitioner for assessment of a swelling in the upper right quadrant. They noted gingival overgrowth associated with partially erupted posterior teeth in the same region.





Figure 1. and Figure 2**.** Firm bony swelling distal to upper right deciduous canine with abnormal shaped deciduous molars

At presentation, the child was asymptomatic but had reported pain in the past.

**Intra-oral examination** revealed a firm bony swelling distal to upper right deciduous canine, as a result of buccal and palatal expansion of the maxillary alveolus only. Abnormal shaped deciduous molars were partially erupted and palatally displaced and the gingiva adjacent to the teeth appeared to be thickened .

The remaining upper dentition appeared normal, as was the complete lower arch.(Figures 1 &2).

### Radiographic findings

Radiographic examination revealed mesial-distal separation of URD and URE with possible bone abnormality and missing upper right premolars.

A cone beam CT scan excluded other causes of delayed eruption and provide baseline information. The report confirmed that UR4 and UR5 are absent. The URD and URE are in situ with no evidence of root resoption. The UR6 is infraoccluded and has an immature apex. There are no supernumerary teeth impeding eruption (Figure 3).

A provisional diagnosis of SOD was made and biopsy was arranged to confirm.

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Figure 3. OPT radiograph showing missing upper right premolars and coronal and axial Cone beam CT images showing bone expansion.

A bone biopsy was performed under general anaesthesia from the right maxillary alveolus, including surrounding gingival tissue.

### Histopathology

Biopsy samples were submitted and sent for short decalcification and immunohistochemical staining. This showed microscopic features of dense fibrocollagenous connective tissue accompanied by a few irregular deposits of immature woven bone (Figure 4).

Histopathology report confirmed the clinical diagnosis of Segmental Odontomaxillary Dysplasia.

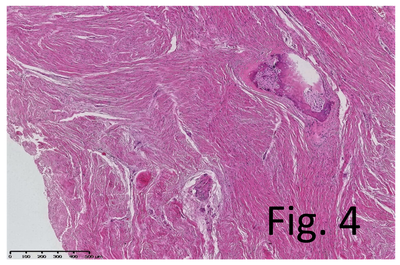



Figure 4: Histopathological features of SOD

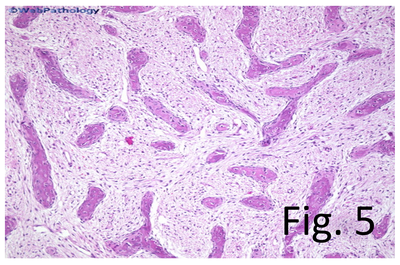



Figure 5: Chinese letters associated with Fibrous dysplasia

### Management

The patient and her parents were reassured that the lesion is benign in nature and that periodic monitoring would be required as delayed eruption of associated permanent teeth is likely. Preventative advice including oral hygiene; fluoride availability and dietary advice was given in accordance to the Delivering Better Oral health toolkit, published by the Department of Health, England.

## DISCUSSION

Differential diagnoses include fibrous dysplasia (FD) and regional odontodysplasia (RO), ghost teeth.

SOD, which differs from FD in that the abnormality is restricted to the alveolus and odontogenic tissues.

FD is not restricted to the alveolus and the microscopic features include “Chinese letters” that represent the irregular trabecular bone (Figure 5).

RO affects the mesenchymal derivatives of the teeth including the enamel, dentine, pulp and dental follicle. RO can present with failure of eruption and radiographically presents with a ghost like appearance of the teeth, dentinal clefts and large pulp chambers.

For the future it is important to carry out regular reviews for this child to monitor eruption of the remaining permanent dentition and management of the absent upper right premolar teeth.

## REFERENCES

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