A mysterious peripheral ossifying fibroma: case report

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ABSTRACT

A case of peripheral ossifying fibroma (POF) in the anterior mandible of a 63-year-old woman is described. The 17 x 23 x 16mm nodular mass was sessile, displaced teeth and had a non-ulcerated, pink surface. Due to the variations in radiographic, clinical and pathological appearance, initially the diagnosis of peripheral odontogenic fibroma was reached. Complex nomenclature and variation in clinical, radiological and pathological appearance of POF and similar masses is discussed.[1]

INTRODUCTION

Peripheral ossifying fibromas (POF) are uncommon, likely reactive gingival overgrowth lesions. They are also known as ossifying fibrous epulis, among several other names. Histopathologically they are formed of mineralised tissue of a varied type: bone, cementum-like material or dystrophic calcification encased in a highly cellular fibroblastic tissue matrix. Predominantly affecting women by the ratio 1.7:1, POFs are most common in the second decade of life in the incisor-cuspid region of the maxilla.[2]

Upon clinical examination, POF present as a single, slow-growing nodular mass occurring on the free gingival margin that can be pedunculated or sessile, with a smooth or ulcerated surface that can vary from pink to red in shade. They can cause tooth displacement,[1] are often asymptomatic, and can vary in size from <1.5cm to 9cm.

Etiology and pathogenesis of POF are unclear: it has features both of a dental anomaly and a neoplasm. It is thought to arise from the periosteal and periodontal membrane as an outcome of irritation from dental plaque, calculus, fixed or non-fixed orthodontic or restorative appliances.[3] In a collation of incidences, the exclusive appearance on the gingiva and presence of oxytalan fibres spread throughout the calcified structures supports the idea of origination from the periodontal ligament. In addition, the reaction of the lesion is cellularly similar to other reactive gingival lesions arising from the periodontal ligament, such as fibrous epulis.[4] Differential diagnoses can vary from peripheral odontogenic fibroma to pyogenic granuloma and fibrous epulis. Due to similar clinical appearances of these lesions histopathological and radiographic correlation are required for definitive diagnosis.

Treatment is with excision, and close follow up is required due to a risk of recurrence that is reported as between 8 to 20%, which is considered high for reactive lesions.[5] Removal of the periodontal ligament and periosteum at the lesion base is required, however tooth extraction is not.

CASE REPORT

A 63-year-old female was referred to the Oral and Maxillofacial Surgery department of the Royal Lancaster Infirmary by her general dental practitioner (GDP) regarding an asymptomatic non-tender, non-ulcerated large bony mass lingual to the lower left central incisor extending to the second premolar, encroaching onto lingual aspects of the teeth. Medically the patient was fit and well, taking no medications and had no allergies. She was a non-smoker, had minimal alcohol intake and volunteered locally after retiring.

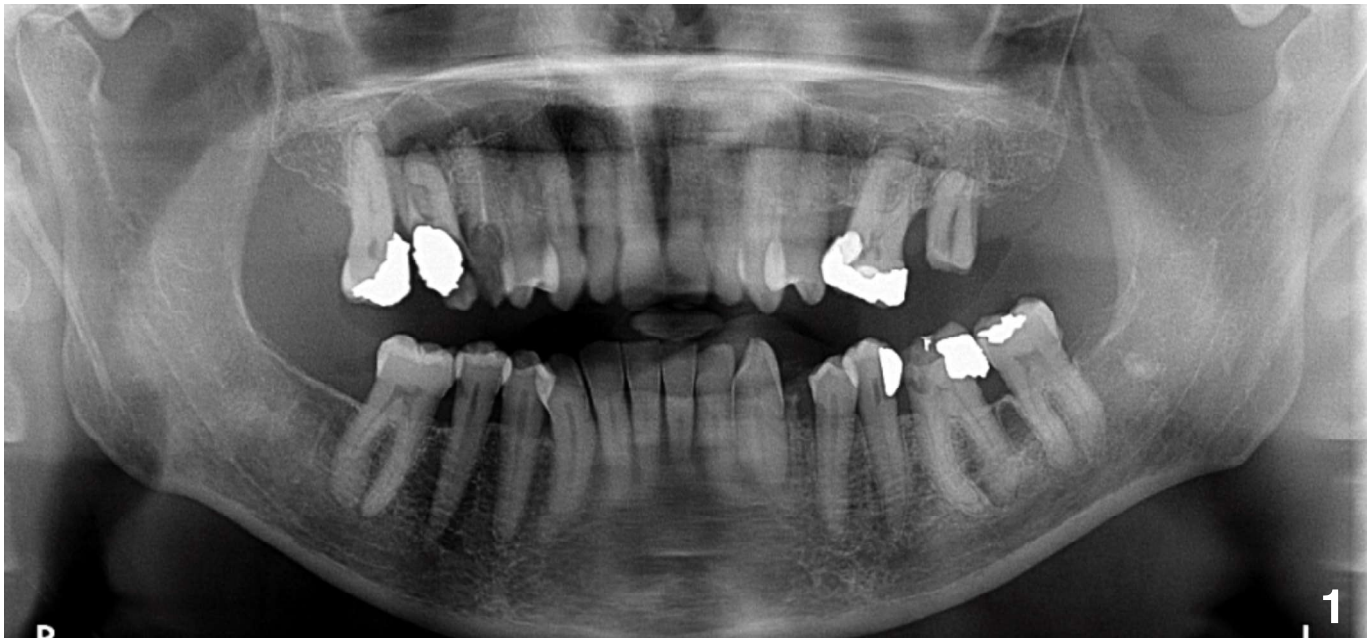
The patient initially presented in July 2019 after referral on a two week wait pathway with a 1.5-2cm lump lingual to LL3/4 causing her teeth and occlusion to feel uneven. She felt this area had enlarged over the previous three months.

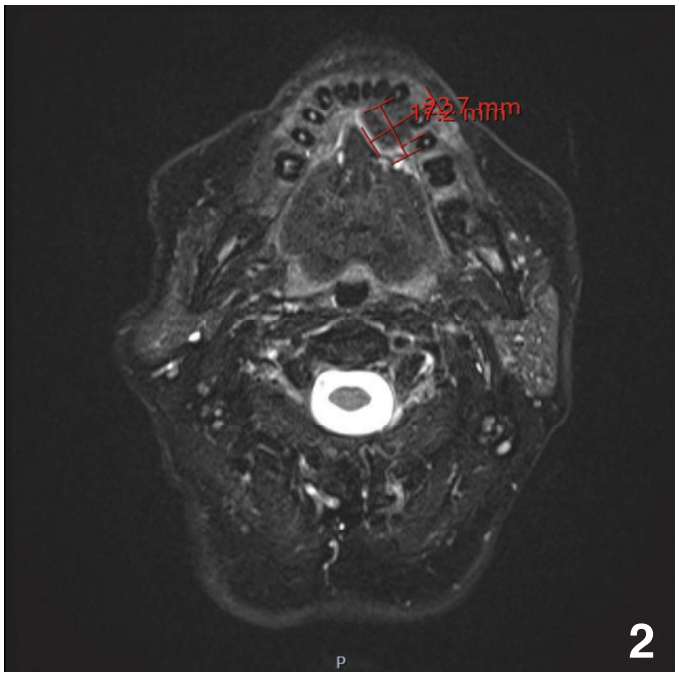
On examination there was a bony mass LL1-LL3/4 encroaching onto the lingual aspect of teeth: this was non-tender and no surface ulceration was present. An orthopantogram (OPG) showed a heavily restored dentition, but no abnormalities. An urgent CT mandible reported that there was a bony excrescence arising from the lingual plate of the mandible adjacent to the socket of the LL3, with no overtly suspicious soft tissue mass. This was diagnosed as a small mandibular torus; the patient was reassured and discharged.

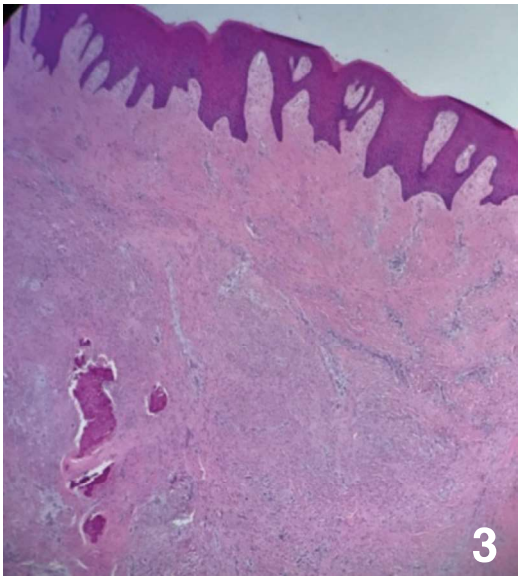
Six months later the patient was re-referred from their GDP, again via the two-week-wait pathway, reporting that the lesion in the left lingual mandible had increased in size. There was no pain, and no further displacement of teeth that she had noticed. On examination there were no lymph nodes palpable, and a 4cm firm soft tissue swelling around from the gingiva lingual to the LL3 with all nearby teeth firm. A large incisional biopsy was taken, an urgent MRI neck with contrast ordered, and an OPG (Fig. 1) which showed presence of a bony spicule between LL3/4. The MRI (Fig. 2) showed a left anterior mandibular gingival mass advising histological confirmation. The biopsy indicated that beneath the epithelium, a mainly fibrotic lesion was present with a small amount of myxoid areas and fragments of apparently dentinoid and calcified tissue. No significant atypia and mitosis were seen. Differential diagnoses were peripheral or central odontogenic fibroma, odontogenic myxofibroma and fibrous epulis.

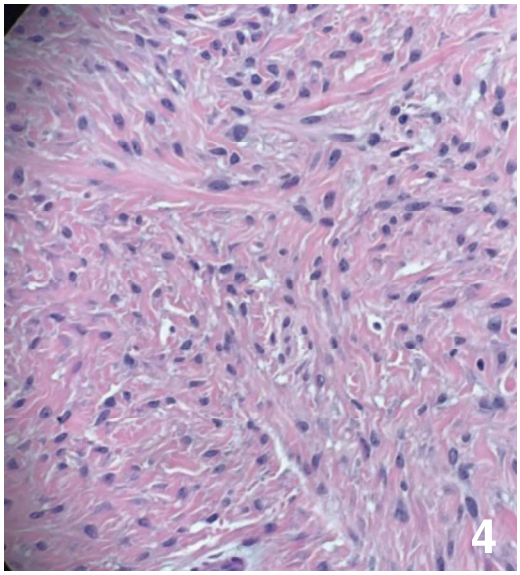
Consent was gained and the patient was listed for excision of this site under general anaesthetic. In surgery in March 2020 this area was removed, including the buccal attached gingiva LR2-LL4. A bony spicule lingual to LL3/4 was smoothed using drill and bur. The area was curetted and cleansed with betadine. A periodontal pack was used to dress the area buccal and lingual of lower anteriors to aid healing, and closure of the sublingual area loosely was achieved with 3-0 resorbable sutures. The soft tissue sample was sent to histology. After one week the patient was reviewed in the outpatient clinic, where she reported feeling well and the area was kept clean. She returned the following week for removal of the periodontal pack under local anaesthetic.

Histology from the site revealed an area composed of irregularly arranged spindle cells with uniform elongated

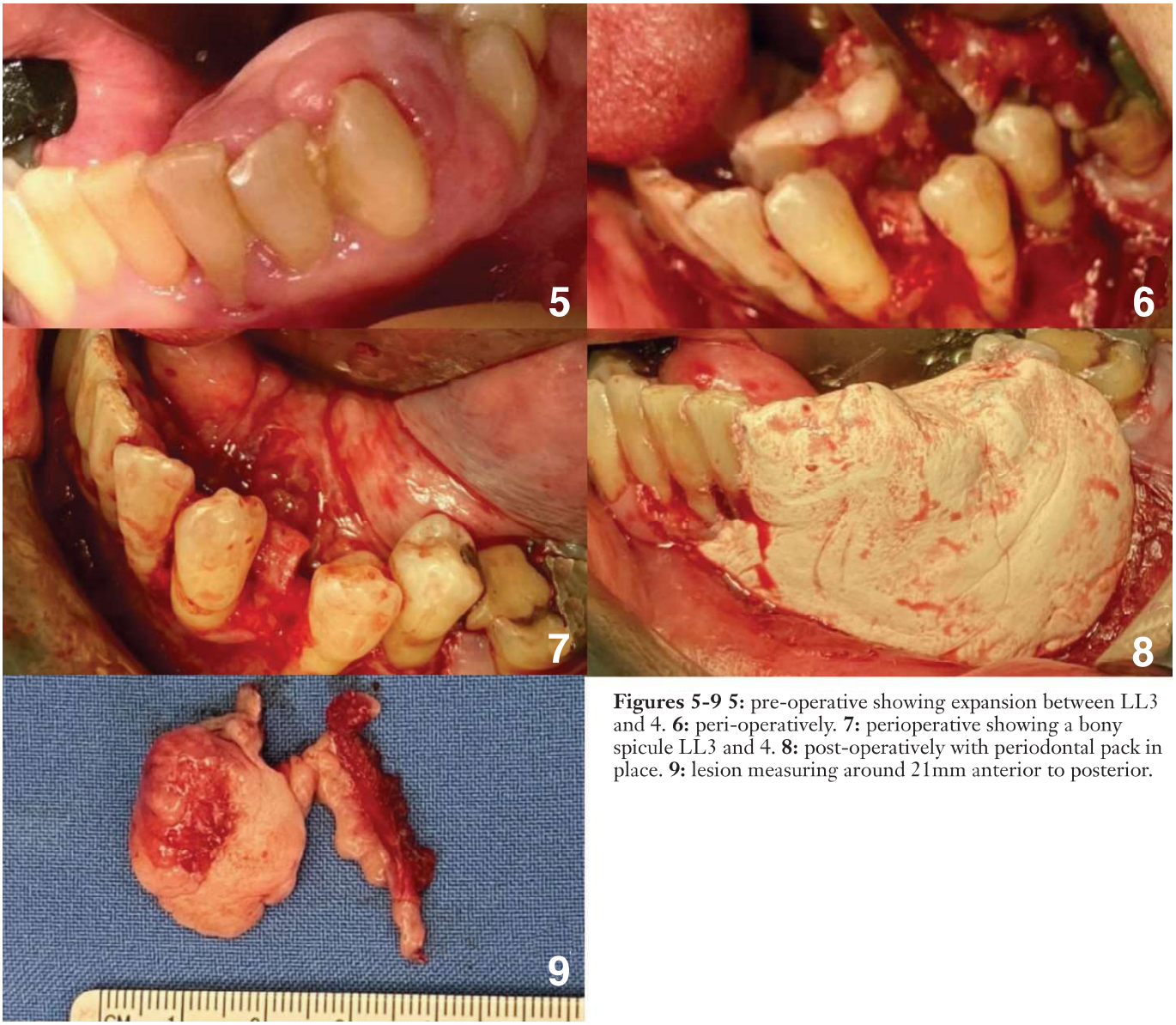








Figures 1-4 1: Orthopantogram taken in January 2020 showing a spur of bone between LL3/LL4. **2:** MRI T2 taken February 2020 showing an area measuring 17 x 23 x 16 mm, arising from the gingiva between LL3 and LL4 with an increased space between these teeth. There is a large sublingual component expanding into the sublingual sulcus. **3:** slide picture showing highly fibroblastic matrix. **4:** slide picture showing epithelium and matrix with embedded areas of osseous material.



Figures 5-9 5: pre-operative showing expansion between LL3 and 4. **6:** peri-operatively. **7:** perioperative showing a bony spicule LL3 and 4. **8:** post-operatively with periodontal pack in place. **9:** lesion measuring around 21mm anterior to posterior.

nuclei, against a background of collagenous stroma with some degree of myxoid change. Focal metaplastic bone formation was noted. These findings favoured an initial diagnosis of peripheral odontogenic fibroma requiring clinical correspondence (Figs 3 and 4). Unfortunately, the sample showed an involved deep margin. After further pathological assessment from Professor Sloan (Professor and Consultant in Cellular Pathology, Newcastle Laboratories), along with radiological and clinical correlation, the mass was defined as a peripheral ossifying fibroma (POF).

Following this histological diagnosis and marginal involvement, follow up will be required every four months with the patient returning sooner if she notices regrowth.

DISCUSSION

There are national and international nomenclature variations with POF, making research and definition in this area difficult and may have added to confusion surrounding these lesions. Commonly used synonyms for POF include peripheral cementifying fibroma; peripheral fibroma with cementogenisis; peripheral fibroma with calcification; calcifying or ossifying fibrous epulis and calcifying fibroblastic granuloma.[6] There have been suggestions that POF are an independent clinical entity instead of a variation of pyogenic granuloma, peripheral giant cell granuloma or reaction fibroma. These three lesions have comparable histological, clinical, sex and site appearance, indicating that they could be inconsistent responses to irritation.[7] POF are thought to arise from the cells of the periodontal ligament, despite there being contrasting views as to whether it is a neoplastic or reactive process. It appears to occur in reaction to irritation from dental plaque, calculus, bacteria or trauma from fixed/non-fixed prostheses or masticatory forces.[6] In this case, there was a small bony spur between LL3/4 – it is possible that the POF developed in reaction to this bony excrescence, or that this spicule was part of the POF's effect on the alveolar bone. They are usually found to be <1.5cm in size, but can be up to six or nine centimetres – the lesion here was around 2.1cm on the lingual aspect of LL2/3/4.

Initial differential diagnoses from the incisional biopsy were peripheral odontogenic fibroma, central odontogenic fibroma and fibrous epulis. Following excision of the lesion, dfferential diagnoses were peripheral odontogenic fibroma (POdF) or odontogenic myxofibroma, favouring POdF. Due to uncertainty, the sample was sent for expert opinion (Professor Sloan, Newcastle Laboratories): this report indicated the diagnoses of POF. This uncertainty along the diagnostic pathway shows the intricate, minute differences these lesions can have, and the importance of clinical, histopathological and radiological correlation. Table 1 aids comparison of peripheral ossifying fibromas to peripheral odontogenic fibromas.

Due to the varied recurrence rates reported as between 8-20%, close follow up is mandatory.

CONCLUSION

This report details an uncommon, benign peripheral ossifying fibroma and its management. Due to non-specific features, this benign tumour was difficult to diagnose both clinically, radiologically and histopathologically. Initially in July 2019 a misdiagnosis of lingual torus was made, meaning that when the patient returned in January 2020 more radical surgery was required to remove the area. Prompt and accurate diagnosis of these likely reactive gingival lesions is essential for correct management and effective patient care. Understanding the intricacies of various odontogenic lesions will allow primary and secondary care practitioners to undertake the correct treatment in a timely manner. Further investigation into the complexities of peripheral odontogenic fibromas would be useful, as well as clarification on the precise nomenclature to be used.

HUMAN ETHICS

Consent was gained from the patient for this case report and clinical photographs.

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Table 1: A comparison between peripheral ossifying fibroma and peripheral odontogenic fibroma.[6, 7]

	Peripheral Ossifying Fibroma (POF)	Peripheral Odontogenic Fibroma (POdF)
Patient	Teenagers/young adults More common in females (1:1.7)	20-40 y/o 2 x more common in females
Features	Soft tissue (gingival) reactive growth nodule Sessile or pedunculated Ulcerated to smooth, regular surface Slow-growing Periodontal origin (debated)	Pedunculated or a sessile firm enlargement with a pink, smooth, nonulcerated mucosal surface on gingiva Close relation to tooth root, crown of UE tooth, site of congenitally missing tooth
Location	Present on interdental gingiva More commonly on anterior maxilla Most commonly related to incisor/canine	Mandible: posterior to first molar Maxilla: anterior to first molar
Histology	No odontogenic epithelium Fibroblasts present Calcifications: bone, cementum-like and dystrophic Likely originates from periodontal ligament	Fibrous tissue with small nests of odontogenic epithelium Can have spindled cells but usually more cellular and fibrocollagenous Calcifications: bone, cementum-like or dytrophic
Treatment	Complete excision	Excision and curettage
Radiology	Rarely shows involvement of the underlying bone Occasionally erosion of superficial bone	Unilocular or multilocular radiolucency Distinct borders ~10% have radiopaque flakes – calcification Can cause tooth displacement and resorption

ACKNOWLEDGEMENT

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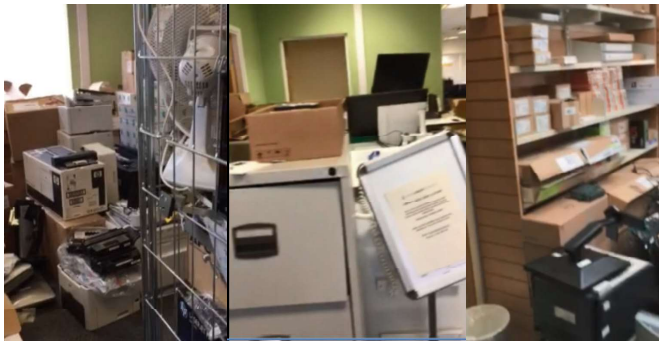
REFERENCES

1. Yadav R, Gulati A. Peripheral ossifying fibroma: a case report. Journal of Oral Science 2009;51(1):151-154. Available from: https://pubmed.ncbi.nlm.nih.gov/19325214/ (accessed 08.07.2020).
2. Buchner A, Hansen L. The histomorphologic spectrum of peripheral ossifying fibroma. Oral Surgery, Oral Medicine, Oral Pathology 1987;63(4):452-461. Available from: https://pubmed.ncbi.nlm.nih.gov/3472146/ (accessed 08.07.2020).
3. Neville B, Damm D, Allen C, Bouquot J. Oral & maxillofacial pathology. 2nd ed. Philadelphia: Saunders; 2002. Soft tissue tumors; pp. 451–2.
4. Mergoni G, Meleti M, Magnolo S, Giovannacci I, Corcione L, Vescovi P. Peripheral ossifying fibroma: A clinicopathologic study of 27 cases and review of the literature with emphasis on histomorphologic features. Journal of Indian Society of Periodontology 2015;19(1):83. Available from: https://www.ncbi.nlm.nih.gov/pmc/articles/PMC4365164/ (accessed 08.07.2020).
5. Cuisia Z, Brannon R. Peripheral ossifying fibroma--a clinical evaluation of 134 pediatric cases. Pediatric Dentistry 2001;23(3):245-8. Available from: https://pubmed.ncbi.nlm.nih.gov/11447957/ (accessed 29.06.2020).
6. Kumar S, Ram S, Jorgensen M, Shuler C, Sedghizadeh P. Multicentric peripheral ossifying fibroma. Journal of Oral Science 2006;48(4):239-243. Available from: https://pubmed.ncbi.nlm.nih.gov/17220623/(accessed 08.07.2020).
7. Kenney J, Kaugars G, Abbey L. Comparison between the peripheral ossifying fibroma and peripheral odontogenic fibroma. Journal of Oral and Maxillofacial Surgery 1989;47(4):378-382. Available from: https://pubmed.ncbi.nlm.nih.gov/2926546/ (accessed 08.07.2020).

COVID-19: How we are delivering library and knowledge services remotely?

Kerry Booth, Library and Knowledge Services, University Hospitals of Morecambe Bay NHS Trust

From Wednesday 18th March 2020 the University Hospitals of Morecambe Bay NHS Foundation Trust (UHMBT) Library and Knowledge Services (LKS) team have been working remotely and operating a reduced service. The library at Barrow (FGH) has remained open with staff being able to access the library collection and computer suite. However, the library space at Lancaster was undergoing a £70,000 refurbishment in March 2020, with the funds provided by NHS England, and as such has not reopened. However, the space has been occupied by the UHMBT I^3 computer engineers who needed a "temporary" new home when their base was reopened as a ward for patients. Hopefully, we will have our site back soon as discussions are underway to relocate I^3. In addition to operating and maintaining the library services remotely, members of the library team have also volunteered to support the community and other UHMBT departments during the pandemic. These placements were located primarily in the scrub exchange and participating in the 'Meals on Wheels' programme.



Still images taken from a video of RLI library space – 16.04.20.

CHANGES TO THE PROVISION OF SERVICE

Some of the services have been affected as a result of the lockdown, the book request and inter library loans (ILLs) service were temporarily suspended; books borrowed from other Trust library services were returned but the LKS team are not able to process any new ILLs.

Whilst library staff are unavailable for face to face enquiries; staff are able to answer enquires using Microsoft Teams and email. The library staff are also available to provide article, literature and information requests as usual and any staff training sessions, such as database searching, have taken place using Teams.

In terms of books out on loan; the decision was taken to renew all items currently borrowed until 1st July; this has been extended until 1st September, therefore staff and students have not needed to renew their loans. Staff and students have been able to return their books to the Barrow library as usual, with Lancaster library calling in a favour with the education centre team, who have agreed to temporarily store any returns.

BUSINESS AS USUAL … REMOTELY

The LKS also continues to support the UHMBT policy team by undertaking the standard corporate document reference checks received, alongside providing information searches for the number of new policies arising as a result of the COVID-19 pandemic.

The LKS team have been heavily involved in the UHMBT Urology Inquiry since September 2019 and this work has continued throughout the lockdown period. The LKS team have been at the centre of information collection for the Inquiry team. Designated library staff