CASE REPORT

Benign cystic mesothelioma of the peritoneum in a 12-year-old boy, diagnosed via telepathology

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SUMMARY

Low resource countries often have inadequate healthcare systems that, among other problems, suffer from a severe shortage of local expertise. In fields such as pathology, however, where diagnoses are mainly based on macroscopic and histological images, telemedicine can provide an opportunity to supplement the local capabilities by involving a world-wide network of experts. For this, the local pathologist can upload images to an online platform and then consult with international colleagues via teleconferencing, which can be particularly useful in rare or difficult cases. We present the case of a 12-year-old Afghan boy with benign cystic mesothelioma, an extremely rare benign tumour. Using the online platform IPath, we were able to diagnose the eighth reported case of this rare condition in a child, in collaboration with our colleagues in Afghanistan.

BACKGROUND

Providing healthcare in low resource countries is a challenging problem. In Afghanistan, three decades of international and civil war have destroyed the local healthcare system. In the past 10 years, international organisations have tried to re-establish healthcare, concentrating on urgent problems such as care for mothers and children, and prevention and treatment of infectious diseases. Medical fields such as pathology, which are not directly involved in patient care, however, are not in the main focus of international organisations. This leads to a lack in local expertise and equipment regarding pathological conditions, making the diagnosis of rare conditions nearly impossible. In the past years, attempts have been made to use telepathology as a tool to overcome these restrictions.¹ We report a case of an exceedingly rare condition, a benign cystic mesothelioma (BCM) in a 12-year-old boy in Afghanistan, diagnosed with the help of telepathology.

CASE PRESENTATION

A 12-year-old Afghan boy was admitted with abdominal pain to the hospital in Mazar-i-Sharif, in Afghanistan. The patient had a history of abdominal pain for 4 years, which had increased in severity in the past month. Ultrasound examination showed a large septated cystic mass occupying the lower abdomen and left flank area (figure 1). Laparotomy was performed. During surgery, a large intra-abdominal mass located under the diaphragm on the left side extending to the pelvis and in contact with the omentum majus, was found and removed. The mass consisted of multiple cystic

Figure 1 Abdominal ultrasound: large septated cystic mass in the lower abdomen and left flank area.

structures, unattached to any organ and with no infiltrating growth.

The material was sent for pathological evaluation to a private pathology lab in Mazar-I-Sharif.

Macroscopic evaluation revealed a mass measuring 20×14×5 cm, with multiple thin-walled cysts (figure 2). Microscopically, multiple cyst-like structures with a cubic or flattened lining could be seen,

Figure 2 Macroscopic picture: large abdominal mass with multiple cystic structures.

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Figure 3 Screenshot of the IPath telepathology platform.

surrounded by soft tissue with prominent inflammation. Cytological atypia was absent and mitotic figures were rare. Advanced procedures to establish a diagnosis were not available on-site; therefore, histological and macroscopic pictures were uploaded by the Afghan pathologist (RR) to a telepathology platform for further evaluation (figure 3). The pictures were revised by a pathologist in Germany (NH), who raised the suspicion of BCM. To verify this further, immunohistochemical staining would have been required, which was not available in Mazar-i-Sharif. A preliminary pathology report with the suspicion of BCM was filed by the Afghan pathologist and the paraffin embedded material was sent to Germany, where immunohistochemistry was performed. The lining of the cystic spaces was positive for calretinin, cytokeratin 5/6 and cytokeratin 7. This confirmed the diagnosis of BCM (figure 4). The result

of the immunohistochemical staining was uploaded on IPath and the Afghan pathologist (RR) sent the concluding pathology report to the surgeon (figure 5).

OUTCOME AND FOLLOW-UP

Six months later, the patient was well, with a small residual tumour mass between spleen and kidney on the left side. He was instructed to return for follow-up within another 6 months.

DISCUSSION

BCM is a rare condition that was first described by Mennemeyer and Smith in 1979.² So far, fewer than 200 cases have been reported,³ most of them in women of reproductive

Figure 4 H&E stain ((A) $\times 12.5$ and (B) $\times 100$) and immunohistochemical staining ((C) Cytokeratin 7, $\times 100$, (D) Calretinin, $\times 400$) of the abdominal mass shows the typical morphology and immunohistochemical staining pattern of benign cystic mesothelioma.

Figure 5 Flow chart of pathological assessment using telepathology.

age. BCM is even rarer in children, especially boys, with only seven cases in the literature.^{4–6}

The lesion has a high recurrence rate and an unspecific clinical presentation, with pain and tenderness or a palpable mass. BCM is considered a benign condition with only a few reported deaths.^{7,8} The aetiology is unknown; it is unclear if the lesion is a true neoplastic process or a reactive condition. It often occurs after endometriosis, chronic infection or surgery, which suggests a reactive aetiology.⁹ Since BCM is very rare and has the capacity to imitate other benign and malignant tumours such as lymphangioma, adenocarcinoma or malignant mesothelioma, it is often difficult to diagnose. The putative reactive aetiology explains the rarity of the entity in children, since endometriosis as a determining factor does not exist and previous operations or chronic infections are uncommon in children.

The diagnosis of rare conditions such as BCM is even more challenging in developing countries, due to the limitation of staining methods such as immunohistochemistry and the lack of local expertise. Since the training of pathologists is time consuming and expensive, low resource countries, including Afghanistan, suffer from a shortage of experienced surgical pathologists to provide adequate service to clinics. Owing to their low numbers, it is further not possible for a pathologist to ask local experts for their opinion, and an established national consulting network for rare cases does not exist. Furthermore, advanced techniques such as immunohistochemistry or special stains are often not established.

In Afghanistan, foreign countries such as the UK and Germany are engaged in rebuilding the healthcare system. Part of this engagement was the reconstruction of the hospital in Mazar-i-Sharif by the German ministry of development, including the equipment for telemedicine, providing the possibility to overcome the lack of human resources and shortage of local experts as well as the shortage of equipment. Additionally, a private pathology lab was established, equipped with the help of donations from German pathologists (figure 6).

Telepathology and telecytology may be effective tools to transfer both knowledge and experience to any place in the world, and to enable, for instance, pathologists in low resource countries to benefit from the experience of experts around the world, especially since telepathology is comparatively simple to establish. A microscope, a digital microscope camera and an internet-enabled computer are all that is needed, with total initial costs of €4100–€4300 (2900–3000 GBP) for the hardware and running costs of €20 per month (14 GBP) for internet access.

Figure 6 The Mazar-i-Sharif pathology lab.

In the field of telepathology, in particular, store-and-forward consultation technologies such as IPath have been gaining momentum in the past years.^{10–12}

IPath is a store-and-forward telepathology service provided by the University of Basel, Switzerland. IPath supports the setup of user-defined groups to which information on cases can be uploaded. Currently, several groups from different countries are active on the platform. Some of the groups are focused on support related to, or discussion of, difficult pathological conditions, while others, such as the Afghanistan group, emphasise basic pathological diagnostics and training of local pathologists.

The Afghanistan IPath group was started in 2010. The aim of the group is to establish a pathological service and pathological training in Mazar-i-Sharif. The local pathologist processes the material and uploads cytological, histological and, if necessary, macroscopic pictures of the material to IPath. International pathologists who have access to the Afghanistan group make their statements online. Usually, a diagnosis can be made based on the pictures provided; sometimes further pictures in higher or lower magnification of specified regions of the slides are requested. In addition, difficult cases can be discussed in a weekly teleconference (via Skype). The major problem in this approach is inadequate sampling of the slide and poor image quality, which can result in wrong diagnoses or the possibility of no diagnosis at all. Diagnostic accuracy increases with adequate training of the local pathologist and constant feedback.

By connecting the pathology lab in Mazar-i-Sharif to an international telepathology network such as IPath, local pathology service and in consequence medical care in general could be improved significantly.

Telemedicine has been used before to enable the correct treatment of patients with rare diseases.^{13,14} In our case, the diagnosis of the 8th reported case of a BCM in a child was only possible with the help of telepathology, leading to correct clinical handling of the patient in the form of surgical therapy and follow-up for recurrences. Our case is an example of how telepathology provides an opportunity to expand the scope of medical care in low resource countries such as Afghanistan.

Learning points

- ▶ Telemedicine can be an important tool to improve medical care in low resource countries.
- ▶ Telepathology is predestined for use in telemedicine due to the shortage of experienced pathologists in most low resource countries and because of the possibility of sending histological and macroscopic pictures, the essential requirements for pathological assessment, via internet. Therefore, it is possible for multiple experts to revise cases without having to be present on-site.
- ▶ With the help of telepathology, even rare and difficult cases can be diagnosed correctly in low resource countries.

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