

Infratemporal fibrolipoma in a 9-year-old female patient

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Summary Fibrolipoma (FL) is a rare subtype of a benign neoplasm as of lipoma, as well as being rarely present in the maxillo-facial area. The reported case is a significant cause for the natural history description of the development of the maxillary sinus immediately adjacent to the lesion was affected. The authors report the case of a female patient, aged 9. In the case reported by us, during a 5-year observation, the development of pediatric fibrolipoma in the base of skull. In addition, after the surgery, orthodontic disturbances developed. Such a course may plead in favour for the necessity of an earlier decision on surgical treatment. Our report is a significant contribution to knowledge of the natural history of FL development the development of pediatric fibrolipoma in the base of skull. It shows that growth of the tumour, though very slow, is progressive and changes regional anatomy and function. Therefore, after the primary diagnosis of FL, surgical treatment without a long follow-up should be strongly considered.

Key words: neoplasms, therapeutics, surgical oncology, pediatrics.

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Background

Fibrolipoma (FL) is a rare subtype of lipoma, a benign soft-tissue neoplasm [1]. Scientific literature analysis shows that FL amounts to 1.6% of all lipomas present in the facial area [2]. The neoplasm is of an extremely benign character, and its development leads to local symptoms. Clinically, it presents with a slowly growing, asymmetrical tumour with mid-soft consistency. The neoplasm may be present in every part of the human body, and within the head area, it is most frequently present in the cheeks, at times within the area of the lips, parotid gland or in the oral cavity. FL is typically built of a mature adipocytes mass surrounded by a thin fibrous capsule [3–5]. The correct diagnosis and implementation of the right course of treatment of FL in the maxillo-facial region is very important [6]. After a total resection, the tumour rarely recurs. Unfortunately, patients often seek medical attention only after they develop alarming symptoms or experience impairment in daily functioning [7].

Case presentation

A female patient, aged 9, was admitted to the Maxillo-Facial Clinic of Pomeranian Medical University in Szczecin with diagnosis of a large tumour localised within the area of left infratemporal fossa. The tumour was detected in the child when she was 2 years old due to the presence of a small bulge of the left cheek, which was associated with pain, diagnosed by the family doctor and referred to a CT scan of the head. Imaging showed an expanding growth of the tumour, localised posteriorly to the posterior wall of the left maxillary sinus. The child was admitted to the Oncology Clinic for Children where a tumour biopsy was performed to determine its character and histopathological type. The results showed the presence of only the hyphae of adipose tissue in the collected material. It was decided then – after an inter-disciplinary consultation with

neurosurgical specialists, children's oncologists and maxillo-facial surgeons – that due to the benign character of tumour, the child should be subjected to constant observation and periodic control.

Longstanding control of the child and annual CT scans of her head did not indicated obvious changes within the area of the tumour; apart from slow, non-significant growth, there were no disorders within the facial skeleton, no pain discomfort, nor disturbances in the stomatognathic system.

After a period of 5 years of continuous control, in MRI and CT scans, there was a heterogenous large solid tumour mass found with the density of fat with hyperdense strands in the left infratemporal fossa (Figures 1 and 2). The tumour size was approximately 66 x 50 x 42 mm; after a contrast, the tumour was not effectively enhanced by the contrast (Figure 3). In the central part of tumour, a non-regular polycyclic calcification was visible (Figures 3 and 4). The tumour fully fulfilled the infratemporal fossa causing compression, modelling and shifting of the adjacent structures; however, there was no sign of infiltration. In the examination, there was a significant decrease in size of the left maxillary sinus in the anterior-posterior dimension due to compression of the tumour mass (Figure 4).

Due to the increase in tumour mass, compression on the adjacent structures, as well as possible risk of irreversible malformations within the area of facial skeletal structures, a decision was made for surgical excision.

The patient was admitted to the Maxillo-Facial Surgery Clinic of the Pomeranian Medical University in Szczecin, where the surgery was performed. With the use of a hemi-coronal incision, extended to the pre-auricular incision on the left side (Figure 5), the temporal muscle was exposed and cut, and the zygomatic arch was cut with a piezo-electric saw (at the two opposite ends) and retracted at the masseter muscle insertion. A tumour mass curettage was performed and was referred to a histopathological examination. There was a plate osteosynthesis of the left zygomatic arch.



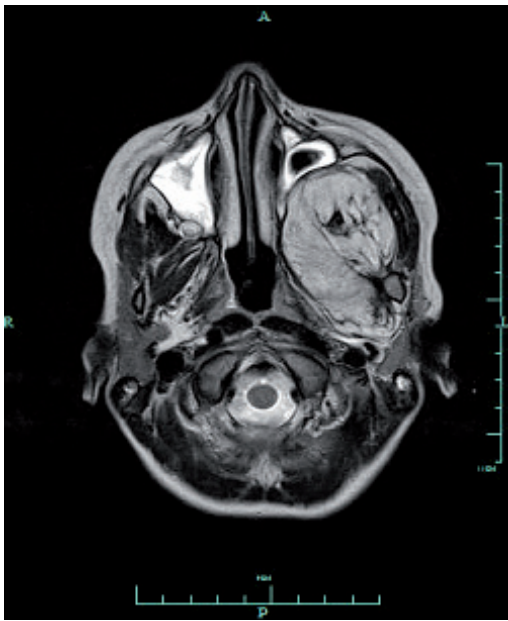


Figure 1. MRI image of the infratemporal fossa tumour on the left side

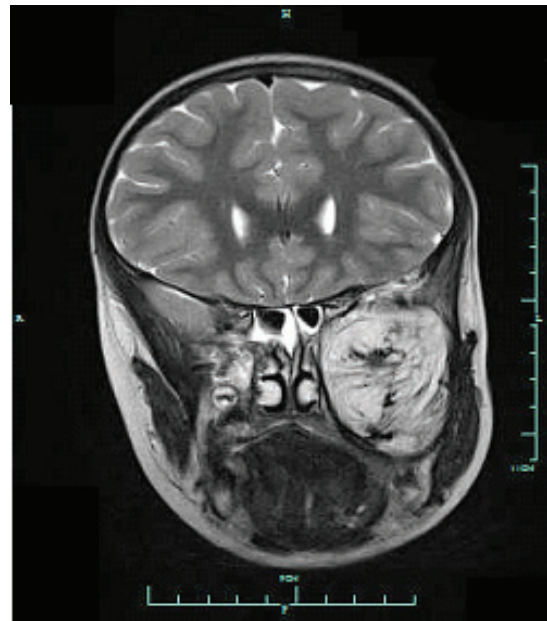


Figure 2. MRI image of tumour (forehead plane)



Figure 3. CT scan image, lack of contrast enhancing within the tumour area

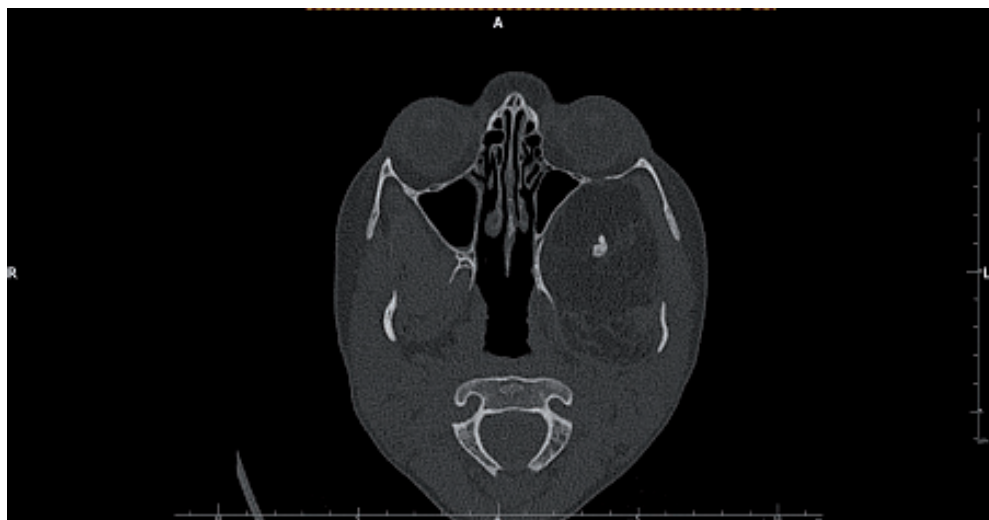


Figure 4. CT scan image of left infratemporal fossa tumour – visible jaw sinuses asymmetry caused by the tumour pressure



Figure 5. Marking of the planned surgical incision (hemicoronal approach)

There were no complications during the post-surgical period. The patient was discharged home with the recommendation of periodic monthly control at the Maxillo-Facial Surgery Clinic for Ambulatory Patients (Figure 6).



Figure 6. Postoperative image after removing sutures

Approximately eight weeks after the surgery, there was a significant limitation in mouth opening found in the patient. The patient was referred to an orthodontic consultation, which showed mouth opening was 8 mm, with a 10 mm skewing of the mandible to the left. A Therabite jaw motion rehabilitation system was applied in the patient. After approximately one month of using the system, on the follow-up appointment at the orthodontist, there was a mouth opening of 30 mm and a 6.5 mm skewing of the mandible to the left (Figure 7).

At present, the patient is under continuous control of the medical team of the Maxillo-Facial Surgery Clinic, Oncological Clinic for Children, as well as the orthodontist (Figure 8).



Figure 7. Mouth opening after orthodontic rehabilitation



Figure 8. Postoperative image after complete surgical wound healing

Discussion and conclusions

Fibrolipoma, classified by the WHO as a lipoma sub-type, is a benign tumour rarely present within the maxillo-facial area. However, if it occurs, it is most frequently present within the buccal tissues and rarely in the lip, parotid gland or tongue. It is most common in patients aged 40 to 60 years [8, 9]. Due to its slow and expanding growth without infiltration or damaging the adjacent structures, frequently without accompanying conditions, it is often unnoticed by the patients, or simply ignored. All the more, its localisation and growth within the area of the infratemporal fossa may occur without any symptoms. To the best of our knowledge, cases of this type of tumour in such a localisation are rarely described in scientific literature and concern mostly adult patients. The authors of the article have never come across a case report on fibrolipoma of the infratemporal fossa in a child in scientific literature [10, 11].

Superficial lipomas may be diagnosed by palpation [6, 12]. Deep lesions require diagnostic imaging, using computed tomography, magnetic resonance or ultrasound [13, 14]. Having adequate information and starting treatment at the right time is important in the case of lipomas occurring in the maxillo-facial region [15]. The basic method of managing benign tumours of this type within the area of the head, as well as other body parts, is surgical treatment [16, 17]. In the case of such tumour localisation in an adult patient, the treatment is not a simple nor frequently performed procedure. It is fraught with a significant mid- and post-surgical risk.

The main mid-surgical risk is the possibility of damaging the upper branch of the facial nerve, which leaves the parotid plexus and runs towards the orbicularis oculi muscle, and the forehead muscles, which may lead to upper eyelid closure disturbance and which may interfere with upper eyelid closure and frowning [18, 19]. Careless surgery may also lead to mandibular artery damage, of which the second part is localised in the infratemporal fossa, as well as damaging the blood vessels of the pterygoid plexus; it may therefore lead to massive and difficult to overcome bleeding, which may be a life threatening condition in a young child.

During the post-surgical period, there may occur some disturbances within the temporo-mandibular joint, associated with constriction of damaged muscles, as well as the occurrence of soft tissues scars; both conditions need intensive post-surgical rehabilitation. In the most extreme form, temporo-mandibular joint ankylosis may occur [20].

In the case of FL in children, there may also be a risk of altered development due to the growing lesion or scarring after its resection [21]. However, there is lack of reports in scientific literature on the surgical treatment of FL in children. Therefore, during prognosis, there is no possibility of using other authors' experience. In the case reported by us, during a 5-year observation, a disturbed

development of the maxillary sinus directly adjacent to the change occurred. In addition, after surgery, orthodontic disturbances developed. Such a course may plead in favour of the necessity of an earlier decision on surgical treatment. Our report is a significant contribution to knowledge of the natural history of FL development the development of pediatric fibrolipoma in the base of skull. It shows that growth of the tumour, though very slow, is progressive and changes the regional anatomy and function. Therefore, after the primary diagnosis of FL, surgical treatment without a long follow-up should be strongly considered.

However, the rare occurrence of a neoplasm within the area, as well as lack of reports on its occurrence in childhood, indicate the need for further observations and the gathering of richer clinical material in multi-centre research to recognise the full clinical course.

Recommendations for the primary care physician regarding early detection of fibrolipoma in children. Recommendations were proposed by the authors based on a review of literature and an analysis of the presented case report. They are not of an expert nature:

- when taking a history, note the time when the lesion appeared and its characteristics – whether it has been growing or not, as well as any accompanying symptoms, like fever or pain;
- examine the child thoroughly and evaluate the lesion;
- if traumatic origin of the lesion is ruled out, the child should be urgently referred to an oncologist, and the necessary diagnostic tests should be requested.

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