A case report and literature review on the imaging manifestations of immature teratoma in fetal oral cavity with concurrent intracranial abnormalities

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Introduction. To explore the imaging characteristics of immature teratoma in fetal oral cavity and enhance the diagnostic and differential diagnostic ability for this condition.
Methods. The clinical, pathological, and imaging features of a case of immature teratoma occurring in a fetus were analyzed. Relevant literature was reviewed to summarize the clinical and imaging characteristics.
Results. Imaging manifestations of fetal oral cavity immature teratoma included: (1) A large solid-cystic mass protruding from the fetal oropharynx, closely related to the maxilla and skull base; (2) CT revealed heterogeneous density within the tumor, with a predominantly cystic appearance and presence of calcifications; (3) MRI demonstrated an irregular solid-cystic mass in the lesion's oropharyngeal region, with low signal intensity on T1WI, internal areas of mixed signal intensity, high signal intensity on T2WI, and high signal intensity on DWI within the solid components. Pathological examination confirmed a Grade III immature teratoma in the fetus.
Conclusion. Immature teratoma occurring in the fetal oral cavity is a rare condition. Understanding its clinical and imaging features can improve the level of diagnostic proficiency for this disease.
Keywords. Immature teratoma; Fetus; Oral cavity; Imaging.

INTRODUCTION

Teratomas originate from abnormal proliferation of germ cells and consist of tissues derived from all three germ layers. They commonly occur in the midline axial organs [1]. In adults, teratomas often arise in the ovaries, while in fetuses, they frequently occur in the sacrococcygeal region [2]. The occurrence of a teratoma in the fetal oral cavity, involving the maxilla and skull base, and concurrent intracranial abnormalities, is extremely rare. Due to the complex composition of teratomas, their imaging manifestations are diverse and challenging to diagnose. Prenatal diagnosis relies primarily on ultrasound examination. However, compared to ultrasound, MRI provides more comprehensive visual information, superior tissue density display, and can reveal the presence of associated malformations. It serves as an important complementary diagnostic tool to ultrasound. Fetal oral cavity teratomas are associated with high morbidity and mortality during the perinatal period, highlighting the importance of early diagnosis.

This study discusses the clinical manifestations, imaging features, diagnosis, and differential diagnosis of immature teratomas occurring in the fetal oral cavity based on relevant literature, aiming to enhance radiologists' understanding of this condition.

1.MATERIALS AND METHODS

1.1 Case Information

Patient:Female, 45 years old. Chief complaint: Amenorrhea for 23+ weeks, fetal abnormality detected for 3 days. Medical history: The patient's last menstrual period was on August 26, 2022, with an estimated due date of June 3, 2023. Menstrual cycles were regular. Irregular prenatal check-ups and no nuchal translucency (NT) examination in early pregnancy. On February 5, 2023, our hospital's ultrasonography showed heterogeneous echoes in the fetal facial area, possibly originating from the oral cavity (measuring 665069mm protruding externally with heterogeneous echoes). On February 6, 2023, our hospital's fetal magnetic resonance imaging (MRI) (Figure 3a) suggested abnormal signals in the fetal oropharyngeal region, possibly originating from the oral cavity, considering a teratoma, and right temporal arachnoid cyst. On February 8, 2023, our hospital's prenatal diagnostic ultrasound indicated a gestational age of 23 weeks. Mass in the fetal oral cavity and cystic echoes in the intracranial temporal region: tumor (teratoma?) and associated arachnoid cyst? (Limited visualization of the fetal lip, equivalent to an approximately 615274mm heterogeneous echo protruding externally, closely related to the intracranial cystic echo, suggesting a connection; a cystic echo with a size of approximately 23*22mm visible in the right intracranial temporal region). No significant medical history, no family history of genetic diseases, congenital defects, or hereditary conditions.

Obstetric history: 2-0-1-1. Gave birth to a healthy male infant in 1999, who died from drowning at the age of 3. Gave birth to a healthy female infant in 2008. One induced abortion.

Laboratory examinations: No abnormalities found.

After understanding the condition, the patient and family requested termination of pregnancy. They agreed to undergo prenatal diagnostic procedures, induction of labor, fetal computed tomography (CT) and MRI examinations, and fetal autopsy.

1.2 Methods

Specimens were fixed in 10% neutral buffered formalin, dissected, and sampled for routine dehydration, paraffin embedding, sectioning at 4μm thickness, hematoxylin and eosin (HE) staining, and microscopic observation.

2. RESULTS

2.1 Surgical procedure

The patient received antispasmodic and sedative treatment. Labor induction was performed using mifepristone and misoprostol, resulting in the delivery of a stillborn female infant (Figure 1) weighing approximately 970g and measuring approximately 30cm in length. A large tumor tissue measuring about 16cm×15cm×8cm protruding externally from the fetal oral cavity was observed, with an irregular shape and soft texture, connected to the palate. The family agreed to send the fetal oral tumor for pathological examination. The induction procedure was successful, and the patient did

not experience any significant discomfort. On the second day after induction, the patient's blood pressure was 140/90 mmHg, and alanine aminotransferase (ALT) was 47 U/L. The patient recovered well after surgery.

Figure 1: The female infant weighed approximately 970g and measured approximately 30cm in length. A large tumor tissue, measuring about 16cm×15cm×8cm, was observed protruding externally from the oral cavity. It was connected to the palate, had an irregular shape, and had a soft texture.

2.2 Fetal Specimen CT and MRI Findings

2.2.1 CT Findings

A large mass of soft tissue was observed within the fetal oral cavity, measuring approximately 6.1x6.1x6.8cm. It partially protruded externally from the oral cavity, with clear boundaries and a visible capsule. The density of the soft tissue mass was uneven, showing cystic and solid changes. The solid portion had homogenous density, while the cystic portion had lower density and exhibited calcification. The lesion was attached to the skull base, palate, oropharynx, and mandible with a broad base. Absorption and destruction of the mandibular bone were visible(Figure 2).

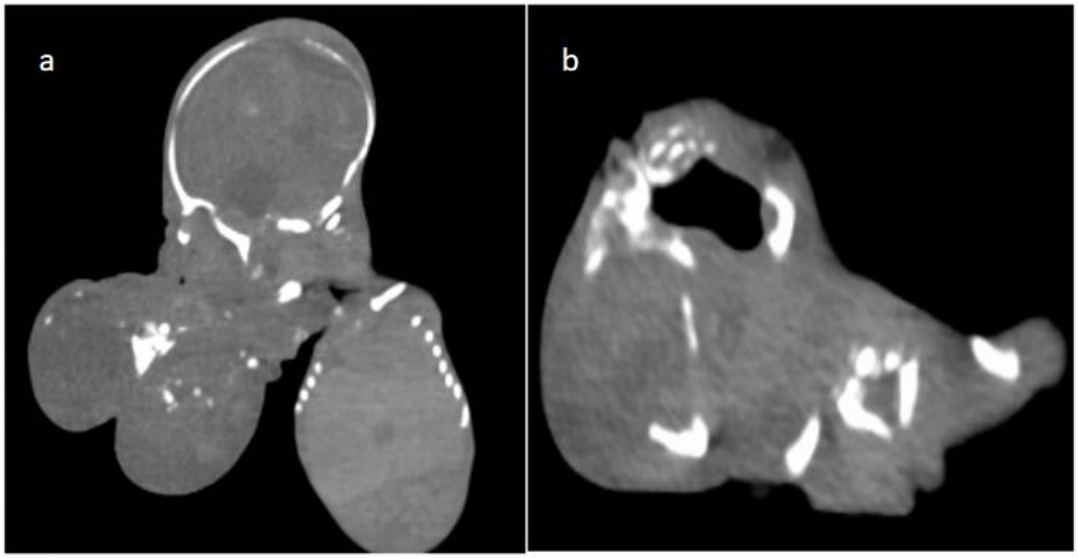

Figure 2 CT Findings: (a) Sagittal view showing a large mass of soft tissue within the fetal oral cavity, measuring approximately 6.1x6.1x6.8cm. It partially protruded externally from the oral cavity with clear boundaries and a visible capsule. The density of the soft tissue mass was uneven, showing cystic and solid changes. The solid portion had homogeneous density, while the cystic portion had lower density and exhibited calcification. The lesion was attached to the skull base, palate, oropharynx, and mandible with a broad base. (b) Transverse view showing bone destruction in the mandible.

2.2.2 MRI Findings

A solid-cystic mass protruding from the fetal facial region was observed, measuring approximately 6.1x6.1x6.8cm. The solid portion showed equal signal intensity on T1-weighted imaging (T1WI) and T2-weighted imaging (T2WI), while the cystic portion appeared as low signal intensity on T1WI and high signal intensity on T2WI. The lesion was attached to the skull base, palate, oropharynx, and mandible with a broad base, most of which protruded externally from the oral cavity. On the right temporal lobe of the fetal brain, a well-defined T2WI hyperintensity, measuring approximately 17x17nm, was visible, causing compression on the adjacent temporal lobe(Figure 3).

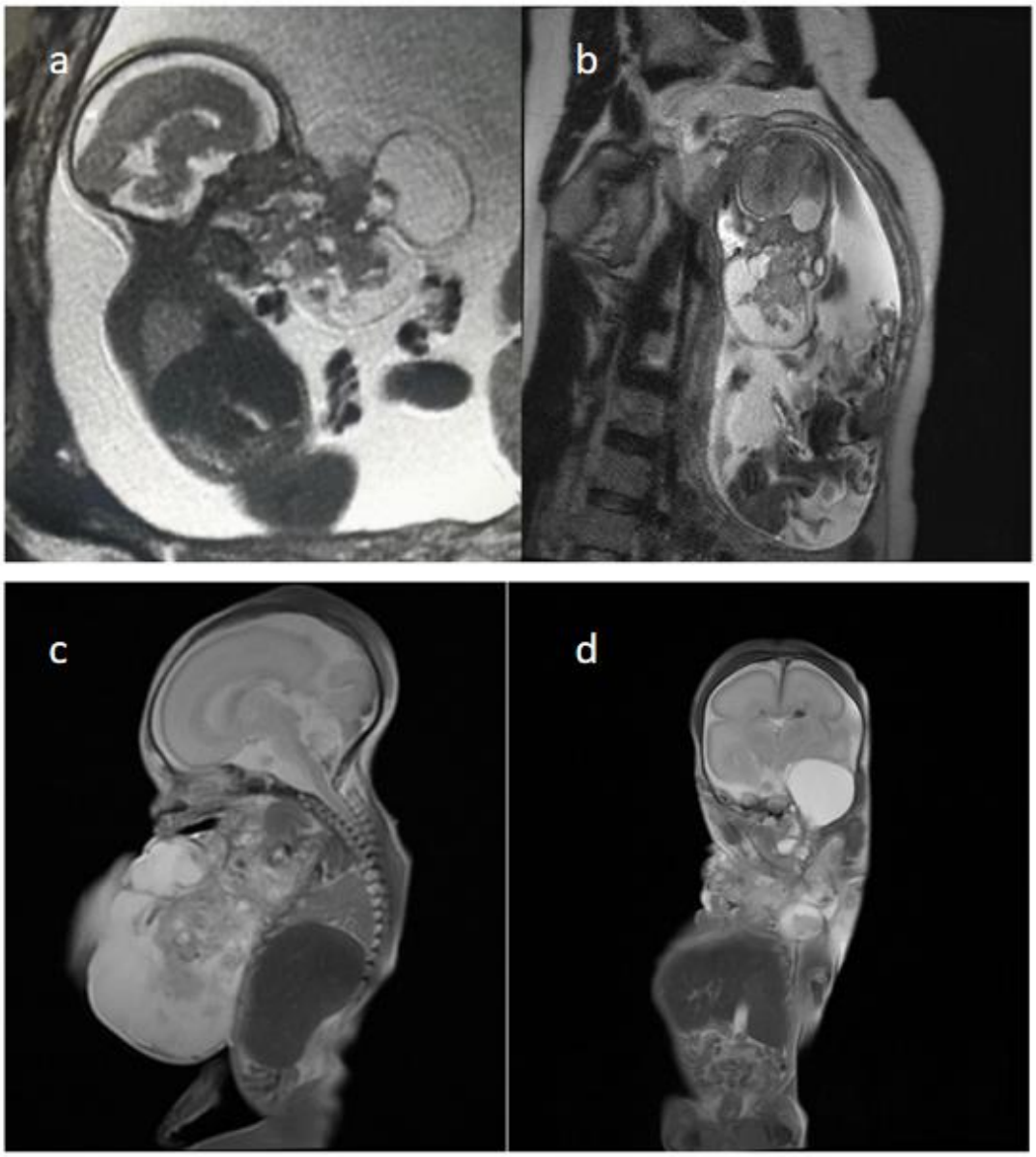

Figure 3 MRI Findings, a-b: Prenatal MRI, Sagittal View (a): A solid-cystic mass protruding from the fetal facial region was observed, measuring approximately 6.1x6.1x6.8cm. The solid portion showed equal signal intensity on both T1-weighted imaging (T1WI) and T2-weighted imaging (T2WI), while the cystic portion appeared as low signal intensity on T1WI and high signal intensity on T2WI. The lesion was attached to the skull base, palate, oropharynx, and mandible with a broad base, most of which protruded externally from the oral cavity. Coronal View (b): A well-defined

T2WI hyperintensity, measuring approximately 17x17nm, was visible in the right temporal lobe of the fetal brain, causing compression on the adjacent temporal lobe.
c-d: Postnatal MRI after induced delivery, Sagittal View (c): The mass could be seen attached to the skull base, palate, oropharynx, and mandible with a broad base, most of which protruded externally from the oral cavity. Coronal View (d): A meningocele of the left temporal lobe was observed.

2.3 Macroscopic Examination
General examination (Figure 4): The fetus measured 25cm×12cm×10cm. There was a protruding mass in the oral cavity, measuring approximately 8×8×7cm. The mass appeared dark red and had an irregular solid-cystic structure. The cystic component contained dark red fluid. The mass was connected to the palate and skull base, and some areas showed firm consistency. No definite organ formation was observed.

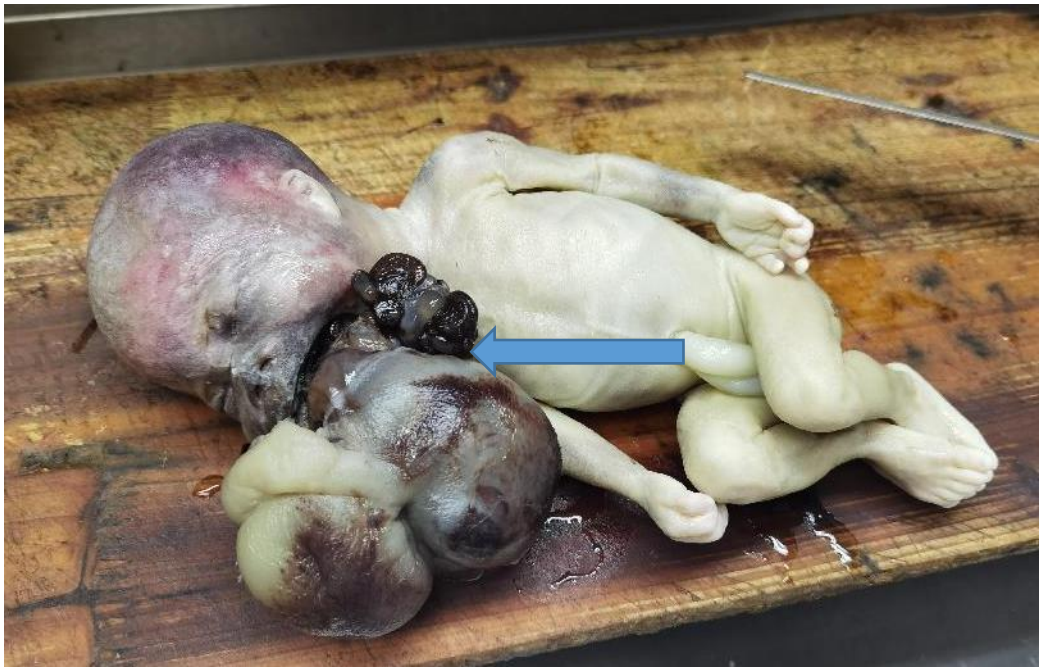

Figure 4 Macroscopic Examination, the fetus measured 25cm × 12cm × 10cm. There was a protruding mass in the oral cavity, measuring approximately 8cm × 8cm × 7cm. The mass appeared dark red and had an irregular solid-cystic structure, with the cyst containing dark red fluid.

2.4 Histopathological Examination

Microscopic observation (Figure 5): The mass consisted of three embryonic tissue layers. Skin appendages, neural glial tissue, choroid plexus, retina, cartilage, smooth muscle, adipose tissue, and vascular tissue were observed. Immature components included primitive neuroepithelium, immature neural glia, immature cartilage, and primitive stromal components, among others.

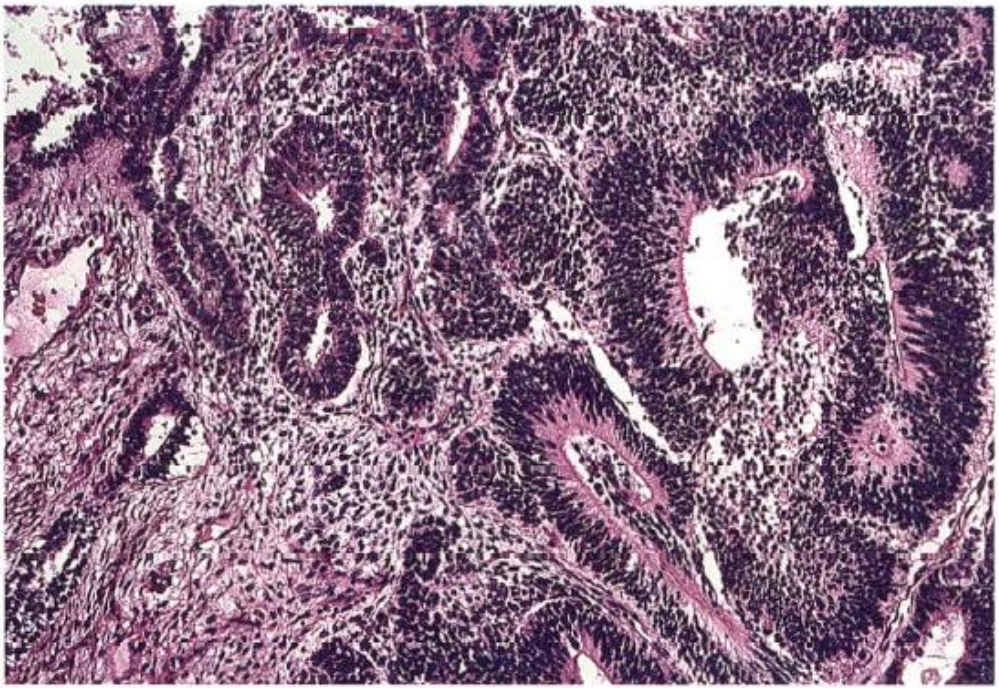

Figure 5 Histopathological Examination

2.5 Pathological Diagnosis

Immature teratoma in the fetus, Grade III. (Grade III diagnostic criteria: Minimal or absent mature tissue; abundant neuroepithelial component, along with a cellular stroma occupying four or more low-power fields in a single slide).

2.6 Follow-up Results

Follow-up at 6 months showed that the mother's condition was good.

DISCUSSION

A teratoma is a tumor derived from germ cells or pluripotent cells of the embryo, primarily occurring in midline axial organs [1], such as the sacrococcygeal region, intracranial region, and sublingual region. Among these, sacrococcygeal teratoma is the most common, with an incidence rate of 1/40,000 to 1/20,000 [2]. Approximately 6%-10% of teratomas are head and neck teratomas, most commonly located in the

neck, while those originating from the oral cavity and connected to the palate and skull base are even rarer. Huang Ying et al. [4] reported a case in which ultrasound misdiagnosed the teratoma as conjoined twin malformation, but it was confirmed to be an oral maxillary teratoma after induced labor. Lai Qiurong et al. [5] and Chen Huiying [6] each reported a case of fetal oral teratoma, both originating from the tongue.

Fetal teratomas are classified as mature and immature. The proportion of immature teratomas is not accurately known. In cases of prenatal diagnosis of teratomas, it has been reported that 61% were diagnosed as mature teratomas, 39% as immature teratomas, and no cases as malignant teratomas [7]. Sacrococcygeal teratomas are mostly benign, while the occurrence of teratomas in other locations is rare, with varying reported proportions. Yoneda et al. [7] reported proportions of 61% and 39% in prenatal diagnosed cases. Liu Xinyou et al. [8] reported that all 12 cases of fetal teratomas were benign. Heerema, McKenney, et al. [9] divided cases based on tumor surgery in utero, tumor discovered in utero but operated on post-birth, and tumor discovered and operated on post-birth into three groups. Group 1 consisted of 100% immature teratomas, group 2 consisted of 71% immature teratomas, and group 3 consisted of 30% immature teratomas.

Immature teratomas often exhibit invasive growth, which can damage surrounding tissues and organs, leading to fetal death [7]. Fetal oral teratomas can block the oral cavity and result in excessive amniotic fluid, causing severe upper respiratory obstruction and high mortality rates [10,11]. Therefore, early detection is crucial. In this case, the immature teratoma originated from the fetal oral cavity, connected to the palate and skull base, protruding outward, exhibiting mobility, and forming a large mass that blocked the oral cavity. This led to upper airway obstruction and excessive amniotic fluid, which may have been the cause of intrauterine fetal death.

Due to the complex composition of teratomas, their imaging characteristics vary, making diagnosis challenging. The pathological diagnosis in this case was an immature grade III teratoma, consisting of tissues from all three germ layers, including skin appendages, neuroglia, choroid plexus, retina, cartilage, smooth muscle, fat, blood vessels, and digestive tract tissue. Among them, the immature components included primitive neuroepithelium, immature neuroglia, immature cartilage, and primitive stromal components, among others. Compared to ultrasound examination, MRI provides a more macroscopic image with better tissue density display, enabling the identification of associated malformations, including those affecting the central nervous system, as well as the determination of airway compression [12]. Combined with ultrasound and magnetic resonance imaging, it is now possible to achieve early prenatal diagnosis of fetal oral teratomas [11,13,14]. In this case, prenatal MRI revealed that the tumor mass was connected to the palate and skull base, exhibiting a large size and protruding into the oral cavity, causing obvious obstruction of the oropharynx and laryngopharynx, and compressing and narrowing the trachea.

Literature reports indicate that approximately 18% of teratomas can be associated with other malformations, such as spina bifida and anencephaly [15]. Therefore, it is important to also consider the development of other organs during diagnosis. In this case, MRI revealed unclear demarcation between the tumor mass and the skull base, along with the presence of an intracranial temporal arachnoid cyst and compression of the adjacent temporal lobe.

Differential diagnosis: When presented with a huge mass originating from the oral cavity, in addition to considering teratoma, one should also consider cervical teratomas, exophthalmos, and facial masses. The former lacks normal lip structure and is located within the oral cavity, while the latter can exhibit lip structures. Furthermore, it is necessary to differentiate from certain intraoral masses such as salivary gland tumors, sublingual cysts, and lymphatic malformations, all of which present as cystic lesions with clear boundaries, rare occurrence of large masses, no significant solid components, and no calcification or fat signals.

Prognosis: Studies have found that cystic teratomas have a better prognosis, while those with a higher tumor volume-to-fetal weight ratio have a poorer prognosis [16]. Some studies have suggested termination of pregnancy for tumors with a diameter greater than 5 cm [17]. Currently, induced labor is generally recommended in China. However, postmortem examination and pathological diagnosis after induced labor are extremely important, as they can confirm the presence of teratomas and determine their benign or malignant nature. In this case, postmortem examination revealed that the tumor affected the palate and skull base, and CT scans revealed destruction of the maxilla and mandible, suggesting a malignant immature teratoma.

In conclusion, fetal oral teratomas are often large in size and can cause obstruction, fetal edema, or obstruct the birth canal during delivery. Therefore, early detection is of crucial importance in determining the fate of the fetus, guiding the mode of delivery, and determining the need for surgical treatment of the fetus and newborn. MRI provides a more macroscopic image with better tissue density display, allowing the identification of associated malformations and greatly assisting in the prenatal diagnosis of fetal oral teratomas.

DATA AVAILABILITY

The simulation experiment data used to support the findings of this study are available from the corresponding author upon request.

CONFLICTS OF INTEREST

The authors declare that there are no conflicts of interest regarding the publication of this paper.

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